UNIVERSITY OF LONDON THESIS

Degree PhD Year 2008 Name of Author OLU SANYA, Bolajoko

Infant Hearing Screening Models for the Early Detection of Permanent Childhood Hearing Loss in Nigeria

BY

Bolajoko Olubukunola Olusanya

July 2007

Institute of Child Health
University College London, UK

ProQuest LLC
789 East Eisenhower Parkway
P.O. Box 1346
Ann Arbor, MI 48106-1346

Declaration

I, Bolajoko Olubunkuola OLUSANYA
confirm that the work presented in this thesis is my own.
Where information has been derived from other sources,
I confirm that this has been so indicated in the thesis.

Signature: **Date**: December 21, 2007.

Motivation

"Just because we cannot see clearly the end of the road,
is no reason for not setting out on an essential journey"

John F. Kennedy

[New York Times, July 2, 1964]

Dedication

To the Almighty,
in whom is hid all the treasures of knowledge,
who has endowed us all with the gift of hearing and
has graciously brought me this far.

Acknowledgements

The author is deeply indebted to Professor Linda Luxon and Professor Sheila Wirz for their painstaking and enthusiastic supervision of this work from inception.

The author is also grateful to several individuals who supported this research at various stages including Dr. Peter Watkin, Siobhan Ryan, Professor David Kemp, Ms Lindsay Kimm, Professor Adrian Davis, Professor Karl White, Dr Randi Winston, Professor Jerry Northern, Professor Robert Ruben, Ms Hanneke Heitling and Professor Keith Sullivan.

The equipment for this study was graciously provided by Natus Medical Inc., USA; Oticon Foundation, Denmark; and Otodynamics (UK) Ltd. The data management software was donated by the National Centre for Hearing Assessment and Management (NCHAM), USA. Training support was received from NHS Newborn Hearing Screening Programme/MRC Hearing and Communication Group, UK. Educational materials for parents and health professionals were sponsored by the Education Trust Fund (ETF), Nigeria. Hearing International Nigeria (HING), a local non-governmental organisation was financial guarantor for the project.

I thank Professor Eyitayo Lambo, the Honourable Federal Minister of Health and Board Chair, Roll Back Malaria Partnership, for officially launching one of the pilot programmes that formed the basis of this research. My gratitude also goes to the Lagos State Health Management Board for approving the use of all hospitals in the State for this pilot programme. I also thank all the Management and Staff of Lagos Island Maternity Hospital, Massey Children's Hospital, Onikan Health Centre, Lagos City Hall Community Health Centre, Sura Primary Health Care Centre, Phonics Hearing Centre, Lagos as well as friends and associates of Hearing International Nigeria for their enthusiastic support for this research.

My husband, Olusegun and children, Oluyemisi, Oluwaseyi and Oluwatobi, were a constant and unfailing source of inspiration without whom this work would not have been contemplated and accomplished.

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List of Key Abbreviations

AABR	-	Automated Auditory Brainstem Response
AAP	-	American Academy of Paediatrics
ABR	-	Auditory Brainstem Response
BCG	-	Bacille Calmette-Guérin (a vaccine for tuberculosis)
CMV	-	Cytomegalovirus
dB HL	-	Decibels Hearing Level
DPOAE	-	Distortion Product Otoacoustic Emissions
DPT	-	Diphtheria, Pertussis and Tetanus
EBT	-	Exchange Blood Transfusion
EHDI	-	Early Hearing Detection and Intervention
EPI	-	Expanded Programme on Immunisation
HVDT	-	Health Visitor Distraction Test
JCIH	-	Joint Committee on Infant Hearing
MDG	-	Millennium Development Goals
NHSP	-	Newborn Hearing Screening Programme, UK
NIH	-	National Institutes of Health, USA
NNJ	-	Neonatal Jaundice
OAE	-	Otoacoustic Emissions
PCEHL	-	Permanent Congenital and Early-onset Hearing Loss
SCBU	-	Special Care Baby Unit
TEOAE	-	Transient Evoked Otoacoustic Emissions
UNESCO	-	United Nations Educational, Scientific and Cultural Organisation
UNHS	-	Universal Newborn (or Neonatal) Hearing Screening
UNICEF	-	United Nations Children's Fund
WBN	-	Well Baby Nursery
WHA	-	World Health Assembly
WHO	-	World Health Organisation

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Glossary of Terms

Acquired Hearing Loss: Reduced hearing sensitivity occurring after birth as a result of injury or disease.

Aural Rehabilitation: Treatment of persons with acquired hearing loss to improve the effectiveness of communication ability, embracing the use of hearing aids, auditory training, speech reading and counselling.

Average Hearing Level: The average of the hearing thresholds in decibels (dB) measured in the better hearing ear at frequencies 500, 1000, 2000 and 4,000 Hz.

Congenital Hearing Loss: Reduced hearing sensitivity existing at or dating from birth resulting from pre- or perinatal pathologic conditions

Critical or Sensitive Phase: Early years of a child's development during which optimal speech and language skills are most readily acquired.

Developing Countries: refer to the 164 countries so classified by the World Health Organisation but are spread from low-income to high-income countries based on World Bank classification.

Early-onset Hearing Loss: Reduced hearing sensitivity occurring within the first 28 days of life.

Hearing Impairment: refers to anatomical defect in hearing and the term is used interchangeably with "hearing loss" in this study. It is conductive when it affects the external or middle ears and sensorineural when it affects the cochlea and the eighth nerve as far as the brain. It is mixed when both the conductive and sensorineural types are involved.

Incidence: The number of new cases of PCEHL in the newborn population during the screening period who have completed the screening protocol.

Infant Hearing Screening: The application of objective hearing tests to babies in the first year of life.

Mild Hearing Loss: Average hearing level of 26 – 40 dB HL. This range is classified as "Slight" or "Grade 1" hearing impairment by WHO.

Minimal Hearing Loss: This term is applied to slight or mild bilateral hearing loss (usually 16 – 40 dB HL) or unilateral hearing loss of any degree of severity.

Moderate Hearing Loss: Average hearing level of 41 – 70 dB HL. This contrasts with WHO's range of 31 – 60 dB HL for children up to 15 years of age and is classified as Grade 2.

Newborn Hearing Screening: The application of objective hearing screening tests to babies at birth or shortly thereafter but not later than the first 28 days of life. It is used interchangeably with "neonatal hearing screening"

Permanent Hearing Loss: Hearing loss with life-long adverse effects on communication and is usually applied to sensorineural hearing loss. Conductive hearing loss can also be permanent if it is related to structural malformation in the middle ear and chronic otitis media. In this study, this term is used synonymously with sensorineural hearing loss.

Prevalence: The number of cases of PCEHL detected in a population of infants during the study period completing the screening protocol.

Profound Hearing Loss: Average hearing level above 90 dB HL in contrast with WHO's range of 81 dB HL and above, or Grade 4.

Protocol: Precise, detailed plan for the administration of a screening test or programme.

Severe Hearing Loss: Average hearing level of 71 – 90 dB HL in contrast with WHO's range of 61 – 80 dB HL or Grade 3.

Slight Hearing Loss: Average hearing level of 16 – 25 dB HL. WHO considers this range as normal or Grade 0.

Yield: The number of cases of PCEHL correctly identified by a screening test or protocol among the subjects enrolled for the study.

Abstract

Permanent congenital and early-onset hearing loss (PCEHL) is associated with significant developmental deficits in speech, language and cognitive skills when detected beyond the first year of life. Hospital-based universal newborn hearing screening is recognised as an essential component of neonatal care worldwide. Although about 32,000 babies are estimated to have PCEHL annually in Nigeria infants are rarely offered any form of hearing screening tests. This research therefore set out to establish suitable infant hearing screening model(s) for Nigeria within the context of the significant proportion of births outside hospital facilities.

In a cross-sectional prospective study, all consecutive newborn babies at the Lagos Island Maternity Hospital, Lagos and babies less than 3 months of age attending four community health centres for BCG immunisation were enrolled over a period of 40 weeks in a two-staged screening protocol consisting of transient evoked otoacoustic emissions (TEOAE) and automated auditory brainstem response (AABR). Those who failed the second-stage screening with AABR were referred for appropriate diagnostic evaluation. Fees were not required of parents for any of the services.

The results demonstrated that both hospital-based and community-based universal infant hearing screening programmes were feasible in Nigeria and that screening tests can be conducted effectively by non-specialists without prior audiological expertise in primary care settings. The screening coverage was over 90%. However, the community-based programme showed a superior yield for children with PCEHL and lower cost-per-baby screened. The only independent predictor of PCEHL under the hospital-based programme was admission into Special Care Baby Unit; while hyperbilirubinaemia necessitating exchange blood transfusion and birth outside hospital facilities were predictive of PCEHL in the community-based programme. Loss to follow-up was the most significant challenge to both programmes while failure to explore likely effects of payments for screening tests on the uptake of services was a major limitation.

Chapter 1

Introduction

1 Introduction

1.1. Overview of Childhood Hearing Impairment

The number of children worldwide with hearing impairment is increasing, and these children face several obstacles in a world in which spoken language is the predominant medium of communication and social interaction. The global estimate for "disabling" (>40 dB HL) hearing impairment by the World Health Organisation (WHO) has more than doubled from 120 million people in 1995 to at least 278 million in 2005, thus confirming hearing impairment as the most prevalent sensory deficit in the human population [WHO, 2006a; Resnikoff et al., 2004; Steel, 2000]. Two-thirds of individuals with hearing impairment live in developing countries, while about 25 per cent represents early childhood-onset hearing loss [WHO, 2006a]. About 2 – 4 babies per 1000 live births in developed countries suffer sensorineural or permanent hearing impairment and this number increases to about 6 per 1000 live births within the neonatal period [Smith, Bale & White, 2005; White, 2004]. Thus, every year about 265,000 to 798, 000 babies worldwide are born with or acquire significant hearing impairment within the neonatal period [Olusanya, Ruben & Parving, 2006].

Lately detected permanent congenital and early-onset hearing loss (PCEHL) is associated with significant and irreversible deficits in linguistic, cognitive and psychosocial development in early childhood [Kennedy et al., 2006; Wake et al., 2004; Moeller, 2000; Yoshinaga–Itano et al., 1998; Hindley, 1997; Robinshaw, 1995 & 1996; McKellin, 1995; Ramkalawan & Davis, 1992; Ross, 1990; Freeman, Malkin & Hastings, 1975; Vermon, 1969]. But if detected and helped with appropriate intervention services within the first year of life children with PCEHL are likely to have comparable development in speech, language and cognitive domains with normal hearing children [Yoshinaga–Itano et al., 1998; Moeller, 2000; Kennedy et al., 2006]. Adequate auditory stimulation in early childhood is therefore an essential

foundation for optimal speech and language development, and for the acquisition of optimal literacy skills [Hannon, 2003; Moore, 2002; Stockard-Pope, 2001; Ruben & Schwartz, 1999; Sininger, Doyle & Moore, 1999; Huttenlocher, 1998; Fisch, 1983].

At school age, PCEHL significantly undermines educational development [Karchmer & Allen, 1999; Bess, Dodd-Murphy & Parker, 1998; Jarvelin et al., 1997; Culbertson & Gilbert, 1986; Allen, 1986; Blair, Peterson & Veihweg, 1985] with substantial lifetime costs to the society [Schroeder et al., 2006; Keren et al., 2002; Mohr et al., 2000]. For example, children with severe or profound hearing impairment by the third grade are likely to be at least 1.5 years behind their normal-hearing peers when enrolled at the same school-age [Allen, 1986]. Although normal hearing children continue to make progress with reading comprehension overtime, the hearing-impaired children make substantially lesser progress even with vigorous educational therapy Figure 1.1 [Marschark, Lang & Albertini, 2002; Allen, 1986].

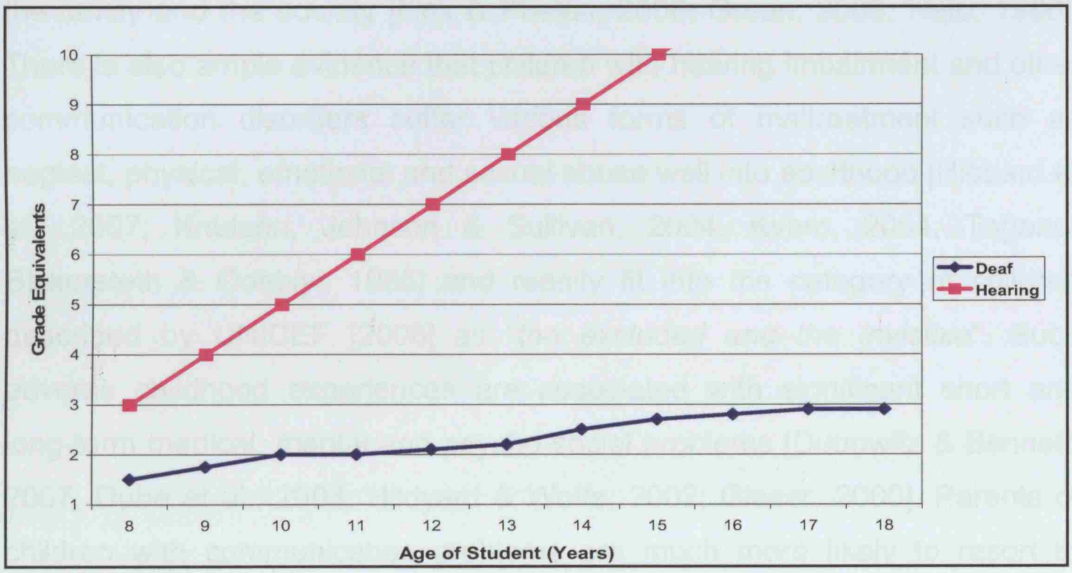


Figure 1.1 Reading Comprehension Scores of Hearing and Deaf Students [Allen, 1986]

Without early language intervention [Spencer & Marschark, 2006] the hearing-impaired children are likely to be about 7.5 years behind their normal-hearing peers on reading comprehension by the age of 15 years [Allen, 1986]. And by the time the children leave school probably at the age of 18 years and over, their reading and comprehension abilities are about 50 - 60% of their chronological age.

This disadvantage continues into adulthood with significant economic and vocational consequences [Ruben, 2000; Jarvelin et al., 1997]. For instance, Ruben [2000] reported that the income for the hearing-impaired population is 40% to 45% lower than what the hearing population earns. Similarly, in the United Kingdom, those with hearing impairment are 3 times more likely to be unemployed than non-disabled persons [Ruben, 2000]. In Denmark, young adults who are hearing-impaired from early childhood are more likely to be trained in crafts than to attain a university education [Parving & Christensen, 1993].

The stigma attached to hearing impairment is of great cost to the individual, the family and the society [Link & Phelan, 2006; Green, 2003; Hetu, 1996]. There is also ample evidence that children with hearing impairment and other communication disorders suffer various forms of maltreatment such as neglect, physical, emotional and sexual abuse well into adulthood [Hibbard et al., 2007; Knutson, Johnson & Sullivan, 2004; Kvam, 2004; Togonu-Bickersteth & Odebiyi, 1985] and readily fit into the category of children described by UNICEF [2006] as "*the excluded and the invisible*". Such adverse childhood experiences are associated with significant short and long-term medical, mental and psycho-social problems [Dubowitz & Bennett, 2007; Dube et al., 2003; Hildyard & Wolfe, 2002; Glaser, 2000]. Parents of children with communication problems are much more likely to resort to physical discipline out of frustration when they perceive a hearing-impaired child's misbehaviours as intentional failure to respond to verbal guidance. While children with disabilities generally experience maltreatment more than children without disabilities, those with communication problems have a

greater preponderance of first incidents from birth to 5 years than any other group of disabled children [Sullivan & Knutson, 2000]. A child's hearing loss is also a source of chronic parenting stress and marital problems [Henggeler et al., 1990; Quittner, Gluekauf & Jackson, 1990; Togonu-Bickersteth & Odebiyi, 1985].

Children with permanent hearing impairment constitute a significant proportion of children with special health care needs who require health and related services of a type or amount beyond that required by children generally [Maulik & Darmstadt, 2007; Couper, 2002; Newacheck et al., 1998]. Although literature on childhood hearing impairment in developing countries is scant, the significantly poorer socio-economic conditions and the fragile healthcare systems in the developing world are likely to raise the burden of hearing impairment well beyond the current evidence from developed countries. Moreover, rehabilitation centres are uncommon and where they have been established they are ill-equipped, poorly run and rarely oriented towards integrating persons with disabilities into the larger society unlike the practice in most of the developed world [Lytle, Johnson & Hui, 2005; Kiyaga & Moores, 2003; Eleweke, 2002; Committee on Nervous System Disorders in Developing Countries, Board on Global Health, 2001; Simeonsson, 1991].

Hearing impairment in early childhood is not readily detectable by routine clinical examination or behavioural observations although it can be suspected by parents through a baby's inattention or erratic response to sound. Depending on the severity, hearing impairment in an infant may not be detected well over 18 months of life by parental suspicion. In fact, all degrees and configurations of permanent hearing impairment in early childhood present with great subtlety and most parents are unable to identify their child's hearing impairment before the associated speech and language delays become apparent [Prendergast, Lartz & Fiedler, 2002; Watkin, Baldwin & Laoide, 1990; Parving, 1984]. In the absence of a systematic effort to screen infants with hearing loss the average age of detection is well over two years and detection may be as late as 6 years [Canale et al., 2006;

Harrison, Roush & Wallace, 2003; Davis et al., 1997; National Institutes of Health (NIH), 1993; Coplan, 1987; Martin et al., 1981]. Parental education is unlikely to fully redress this limitation [Borgstein & Raglan, 1998].

The development of objective, valid, reliable, simple, safe and automated screening technologies consisting of otoacoustic emissions (OAE) and automated auditory brainstem response (AABR) has made the testing of infants achievable from birth for the prompt detection of PCEHL. Consequently, the identification of infants with PCEHL within the first 3 months of life coupled with the provision of appropriate intervention services by 6 months is now commonly considered ideal practice for optimal speech and language development in early childhood [Joint Committee on Infant Hearing (JCIH), 2000; NIH, 1993]. Davies et al [1997] suggested that the most opportune time for detection hearing impairment in children was at birth, at a developmental check in the first year of life, or at school entry. However, from a practical standpoint, particularly in countries where majority of babies are delivered in hospital facilities, screening at birth has the unique advantage of a captive population for achieving the highest possible coverage for the early detection of infants with congenital hearing loss although it would miss infants with early-onset hearing loss. Universal newborn hearing screening (UNHS) before hospital discharge has therefore emerged as a key component of neonatal care in the developed world. [Kolski et al., 2007; Leveque et al., 2007; Neumann et al., 2006; Weichbold, Nekahm-Heis & Welzl-Mueller, 2006; Tsuchiya et al., 2006; Bamford, Uus & Davis, 2005; Pastorino et al., 2005; Flynn et al., 2004; White, 2003; Russ et al., 2002].

Concerned at the growing problem of largely preventable hearing impairment particularly in the developing world and recognising that it constitutes a particularly serious obstacle to optimal development and education, including language acquisition and communication skills, the World Health Assembly (WHA) passed a resolution in May 1995 urging Member States to "prepare national plans for the prevention and control of major causes of avoidable

hearing loss, and for the early detection in babies, toddlers, and children, as well as in the elderly, within the framework of primary health care" [WHO,1995]. While some of the risk factors or causes of hearing impairment are preventable through improved maternal and child care, immunisation coverage and personal/environmental hygiene, the prevailing socio-economic conditions and weak health care systems in many developing countries make sole reliance on primary prevention ineffective or inadequate. Hence, secondary prevention through early detection and management of hearing impairment has become imperative to provide safety-nets for those unavoidably affected [Smith, 2003; Alberti, 1996; Gell et al., 1992; Simeonsson, 1991].

It is pertinent to observe that while the WHA resolution preceded the widespread implementation of UNHS in developed countries and acknowledged the need for early hearing detection, it did not specify modalities for achieving this goal particularly in a developing country such as Nigeria. This was largely because there was no consensus on the best approaches for infant hearing screening internationally at that time [Bess & Paradise, 1994; Gell et al., 1992]. Similarly, although the current WHO strategic directions for maternal and child health in the developing world acknowledge the need for the detection and management of hearing impairment in the first year of life, no specific approaches have been proposed for this purpose [WHO, 2003]. Individual countries have then been left with the challenge of determining possible and culturally-appropriate options for early detection of childhood hearing impairment in general and PCEHL in particular, in their respective populations. The importance of contextualising service delivery for people with communication disorders in the developing world has been well articulated by Hartley & Wirz [2002].

Pilot projects on infant hearing screening have been reported in a growing number of developing countries including Brazil, India, Jordan, Malaysia, Mexico, Oman, Pakistan, Philippines, Saudi Arabia and South Africa attesting to the feasibility of early hearing detection programmes in the developing

world [Olusanya et al., 2007]. However, no such project has been undertaken in Nigeria.

1.2. Background Information on Nigeria

1.2.1. Historical and Socio-demographic Profile

Nigeria is located on the Gulf of Guinea in West Africa and situated between longitudes 3 and 14 degrees east and latitudes 4 and14 degrees north (Figure 1.2). It is bordered on the West by Benin Republic, on the East by Cameroon and on the North by Niger and Chad. The terrain consists of southern lowlands merging into central hills and plateaus; mountains in the southeast and plains in the north; while the climate varies from equatorial in the south, tropical in the centre and arid in the north.

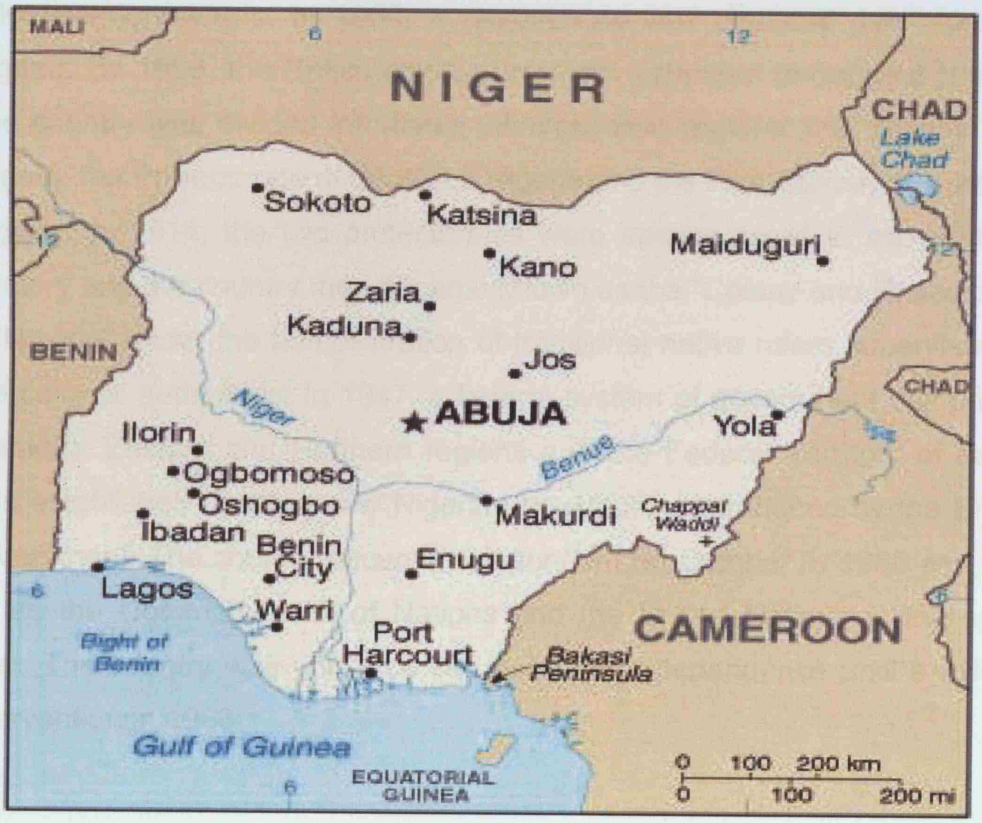


Figure 1.2. Map of Nigeria

Nigeria is the most populous African nation with an estimated population of 140 million people spread on a total land mass of 923, 768 square kilometres. The Rivers Niger and Benue divide the country into three main sections occupied by the dominant ethnic groups: Hausas/Fulanis in the North, the Yorubas in the South-West and the Igbos in the South-East. The Northern Hausa/Fulanis are predominantly Moslems while the Yorubas and Igbos are Christians. Altogether, there are about 374 identifiable ethnic/linguistic groups spread over 774 Local Government Areas (LGAs) in six geopolitical regions: North-West, North-Central, North-East, South-West, South-East and South-South. The Nigerian populace is predominantly rural and approximately one-third live in urban areas.

The emergence of Nigeria as a nation has a strong historical link with the United Kingdom. After the abolition of slave trade in the 19th century (1833), the coastal region of Lagos became a British colony in 1861 serving as a springboard for the subsequent expansion of British trade and political influence northwards. In 1900, a protectorate was declared over northern Nigeria. By 1906, the British colonial rule was extended throughout Nigeria. The country was divided into three administrative regions: the "Colony" (i.e. Lagos), the Protectorate of Southern Nigeria and the Protectorate of Northern Nigeria. In 1914, the two protectorates were amalgamated to into a single territory and the country then became known as the "Colony and Protectorate of Nigeria" under the administration of traditional native rulers supervised by the colonial authorities. In 1947, a federal system of government comprising Western, Eastern and Northern regions and the Federal Territory of Lagos was established under a new Nigerian constitution introduced by the British government. The country became independent on October 1, 1960 and also joined the Commonwealth of Nations and the United Nations in the same year. The country was under civilian rule from independence until a military intervention in 1966.

The period between 1966 and 1979 was characterised by military coups and civil war, followed by a brief return to civilian rule from 1979 to 1983. Between 1983 and 1999 the country again witnessed several military interventions and the longest military rule since independence before the inauguration of the present civilian administration on May 29, 1999 under a new 1999 constitution. The constitution provides for an executive branch headed by a president to be elected on a four year term and a bicameral legislature consisting of a 360-seat house of representatives (or lower chamber) and a 109-seat senate (or upper chamber) equally elected on a four year term. The country is now divided into 36 states with its federal capital in Abuja. Lagos which was the first capital city under the colonial rule, officially ceased to be the capital in December 1991, but has remained the commercial nerve centre for the country.

1.2.2. Economic and Educational Profile

Nigerian economy is historically agrarian and over half of the workforce is still engaged in largely subsistence farming. Major agricultural exports include cash crops such as cocoa and rubber from the south and cotton from the north. Crude oil is the leading mineral produced in Nigeria and it is mostly found in the Niger delta, in the southern part of the country. Petroleum production on an appreciable scale started in the late 1950s and by the early 1970s emerged as the leading earner of foreign revenue. The rapid growth of the petroleum industry since early 1980s has fuelled massive rural-urban migration to the detriment of agricultural production which has ceased to be a major foreign exchange earner for the country. Nigeria's leading trading partners are United Kingdom, USA, Japan, Germany, Netherlands, China, France and Brazil.

Nigeria is the second largest economy in Africa and accounts for 41% of the region's Gross Domestic Product (GDP) [The World Bank, 2006]. The GDP per capita as at 2005 is estimated at US$ 678 (about £400). There was a steady rise in GDP per capita for four consecutive years (1998 – 2001) until 2002 when the growth rate declined from 4.21 per cent in 2001 to 3.27 per cent in 2004. Agriculture remains the major contributor to GDP, currently about 40%. However, Nigeria's enormous potential to generate foreign exchange earnings from its vast reserves of mineral, natural and human resources remains largely untapped as crude oil exports account for about 99 per cent of total export earnings, 85 percent of government revenues, and about 52 percent of GDP. This monolithic revenue base has made the country's economy highly vulnerable to the uncertainties in the world oil market. As crude oil prices fluctuated so did the national revenue, necessitating structural adjustments in public sector spending and recourse to external borrowing which exceeded US$ 30billion by late 1990. Nigeria therefore emerged as one of the most indebted nations in the developing world until end of 2006 when all the major foreign debts were fully repaid.

Political instability and poor management of the economy for many years has resulted in the country's profile as a poor country with 52% of its population living on less than US$1.0 (about 50p) per day. However, since 1999, the civilian administration has embarked on several socio-economic reforms to set the country on the path of steady, real and sustainable growth after many years of stagnation. These reforms include unprecedented privatisation of public enterprises and anti-corruption initiatives which have attracted massive inflow of foreign investments in the last six years. Remarkable progress has also been recorded in the telecommunication and financial services industries. Non-oil growth accelerated to 8.2% in 2005 and is estimated at about 8.9% in 2006. This trend signals less direct involvement of government in the provision of services across all sectors thus offering increasing opportunities for private-sector initiatives.

The adult literacy rate between 2000 and 2004 is reported as 66.8% [WHO, 2006b]. Net primary school enrolment between 1998 and 2004 is estimated as 74% among males and 60% for females. Reforms in the educational sector have resulted in the emergence of several private institutions from kindergarten to university level predominantly in large urban centres across the country to meet the growing demand for education.

1.2.3. Structure and Development of Healthcare Services

The healthcare system has passed through many phases of development and has been greatly shaped by the political history and prevailing socio-economic conditions at each stage. Western or "modern" healthcare services were first introduced to Nigeria by the early European explorers primarily to serve their personal health needs. The services were later extended to the general public as part of disease containment strategy but limited to major urban centres where the Europeans settled. Consequently, the Western healthcare system is widely regarded as a colonial legacy to serve the urban population or the elite, while the rural communities depend largely on traditional medicines dating back from the pre-colonial era [Alubo, 2001]. The only exception was probably the provision of medical care by Christian missionaries through church-owned health facilities largely concentrated in the non-Moslem and some rural communities.

Since the country's independence in 1960, federal, state and local governments have been charged with the responsibility of providing health services organised within a three-tier pyramidal healthcare system. The first level consists of basic primary healthcare largely provided by local governments through primary healthcare centres coordinated by the State Ministry of Health. Several private providers of healthcare also operate at this level. This is followed by the secondary level of care through general hospitals under the responsibility of the state government. Support services such as laboratory, diagnostic, blood bank, rehabilitation and physiotherapy are provided at this level. At the apex are tertiary health institutions which provide

highly specialised referral services for the primary and secondary levels and are usually managed by the federal and state governments. Tertiary services are provided by teaching hospitals and specialist hospitals for specific diseases such as orthopaedic, ophthalmology, psychiatric, obstetric and paediatric cases. The primary health care is the fulcrum of the national health policy and covers services such as health education, reproductive health including family planning, and childhood immunisation. Nigeria is one of the few countries in the developing world to have systematically decentralized the delivery of basic health and education to locally elected administrations [Gupta, Gauri & Khemani, 2004].

The far-reaching structural adjustments since the mid-1980s have led to significant government divestment from educational, social and healthcare services. For instance, public health expenditure as a proportion of federal government budget shrank from an average of 3.5% in the early 1970s to less than 2% in the 1990s [Ogunbekun, Ogunbekun & Orobaton, 1999]. In fact the overall spending on health services in 1991 was only 44% of the value in 1981 with major implications for public hospitals which relied almost entirely on government subventions. This resulted in the near-collapse of acute hospital services due to frequent drug shortages, poorly maintained equipment/facilities and declining staff morale which unfortunately have persisted till date [Chukwuani et al., 2006; Ehiri et al., 2005]. The emergence of private medical enterprise and recourse to traditional medicine became inevitable to meet the healthcare needs of the population which grew at an annual rate of 3 percent. Private health services are delivered by hospitals, clinics, maternity homes, diagnostic centres, and pharmaceutical/patent medicine stores. Traditional medicine refers to health practices, approaches, knowledge and beliefs incorporating plant, animal and mineral based medicines, spiritual therapies, manual techniques and exercises, applied singularly or in combination to treat, diagnose and prevent or maintain well-being [WHO, 2002a]. A corollary of traditional medicine in developed countries is "alternative", "complementary" or "non-conventional" medicine as distinct from modern or orthodox medicine.

Traditional healers offer services such as obstetric care, including family planning, treatment for common illnesses such as malaria, fever, tuberculosis and HIV/AIDS and care for those with mental or neurological disorders and hearing impairment [Andrade & Ross, 2005; Lasisi & Ajuwon, 2002; WHO, 2002a; Kale, 1995; Odebiyi & Togonu-Bickersteth, 1987]. It is not uncommon for people to switch between modern healthcare and traditional medicine or combine both therapies for long-lasting or intractable illnesses and health conditions [Okolocha et al., 1998; Nnadi & Kabat, 1984]. On occasions when the perceived cause of an ailment is an "evil force" or linked to some superstitious beliefs, people more frequently resort to either traditional medicine or "spiritual healing" [Etuk, Itam & Asuquo, 1999]. The integration of traditional medicine with Western medicine remains a controversial subject especially as there are no enforceable standards for traditional practices or legal remedies against malpractices [Orwa, 2007; Ajai, 1990; Tahzib & Daniel, 1986].

As in many other developing countries the private sector altogether, now accounts for the larger share of healthcare expenditure in Nigeria. For instance, government contribution to national health expenditure in developing countries rarely exceed 50% and is as low as 26% in Nigeria and 25% in India compared to an average of 45% in USA, 68% in Australia, 70% in Canada, 80% in Europe and 81% in Japan [Olusanya et al., 2007; WHO, 2006b]. Although private health expenditure accounted for 55% of the total expenditure in USA, only 24% of this amount is attributed to out-of-pocket spending - in sharp contrast to India or Nigeria where out-of-pocket spending accounts for at least 90% of private health expenditure. In effect, out-of-pocket spending, as a percentage of total expenditure, accounts for 76% in Nigeria or 75% in India compared with about 13% in USA or 10% in Europe. Although, out-of-pocket spending is viewed as inequitable as it offers access to health services based on the "ability-to-pay", it remains the pivot of health financing in developing countries. In line with the privatisation policy of the government, it is unlikely that this trend would change in Nigeria. Rather, government is more likely to foster the growth of public-private partnerships in the provision

of health services and the successful implementation of the recently introduced national health insurance scheme (NHIS).

Like many other developing countries, poor nutrition, over-crowding, limited access to safe water, shared toilets and poor hygiene still characterize the living conditions in Nigeria. The healthcare system continually faces the challenge of overwhelming burden of infectious and deadly diseases such as malaria, diphtheria, tetanus, meningococcal meningitis and more recently HIV/AIDS. With infant mortality rate of about 101 and under-5 mortality rate of about 197 per thousand live births in 2004, Nigeria is one of the six countries that account for half of global child deaths [Black, Morris & Bruce, 2003] and it also has the highest prevalence of developmentally disadvantaged children in the developing world and globally [Grantham-McGregor et al., 2007].

However, several global health initiatives such as the Millennium Development Project spearheaded by the UN, the World Bank, WHO, UNICEF and donor organisations are currently seeking to address some of these health challenges in developing countries [United Nations, 2005]. While current global priorities are still largely skewed towards communicable and fatal diseases, there is a growing interest in chronic and non-communicable diseases which generically incorporate all sensory disorders and congenital abnormalities [Fuster & Voûte, 2005; WHO, 2005a; Yach et al., 2005; Horton, 2005]. This global awareness is rapidly extending to childhood hearing impairment particularly as it affects the attainment of the Millennium Development Goals [Lancet, 2007; Olusanya, Ruben & Parving, 2006].

1.3. Rationale for this Study

Based on the projected prevalence of 6 per 1000 in developing countries, about 31,800 babies, of the 5.3 million babies born annually in Nigeria [UNICEF, 2006], are likely to have significant permanent hearing impairment compared with 840 in the UK [Davis et al., 1997]. Less than 40% of these babies are born in established hospitals compared to almost 100% in the UK [UNICEF, 2006]. Besides, there are about 8.7 million hearing-impaired children below the age of 18 years in Nigeria compared with about 30,000 in the UK [Royal National Institutes for the Deaf (RNID), 2003; National Ear Care Programme (NECP), 2002]. Due to the lack of any routine or systematic childhood hearing screening programme in Nigeria parental suspicion is often the primary mode of detection at a mean age of 22 months which is not ideal for effective intervention in all crucial domains of early childhood development [Olusanya, Luxon & Wirz, 2005a]. Enrolment in the schools for the deaf at a mean age of 10.3 years presently constitutes the only intervention option for parents. No programmes exist to facilitate the development of speech and language or optimise the acquisition of literacy skills. By the time these children leave school probably at the age of 18 years and over, their reading and comprehension abilities are about 50-60% of their chronological age. Besides, little or no attention is devoted to the special health and educational needs of these children and their parents which in turn make them vulnerable **to** neglect and various forms of maltreatment.

However, the Federal Ministry of Health in Nigeria is determined to redress this trend in line with the WHA resolution [WHA]. Consequently, the revised National Health Policy now provides for early hearing detection and intervention initiatives, but there is no relevant data currently, to guide the implementation of this policy or serve as baseline for future work [Federal Republic of Nigeria, 2004]. Hence, the urgent need for such a data in a country that accounts for a significant proportion of global burden of infant mortality and developmental disabilities [Grantham-McGregor et al., 2007; Lawn, Cousens & Zupan, 2005; Black, Morris & Bryce, 2003].

The focus of this research therefore, is to determine effective and culturally-appropriate screening model(s) for the early detection of PCEHL in Nigeria as a first and crucial step towards developing support services for the affected children and their families. In view of the prevailing birthing profile in the country, two simultaneous pilot programmes were considered necessary. The first programme consisted of a hospital-based universal newborn hearing screening as currently practised in many developed, and an increasingly number of developing countries while the second programme/model entailed a community-based universal infant hearing screening to cater for the higher percentage of children born outside hospitals.

1.3.1. Overall Study Objective

The purpose of this study is to explore the feasibility of hospital- and community-based universal hearing-screening for infants within the context of primary health care service in Nigeria.

1.3.2. Specific Objectives

- To ascertain the feasibility and effectiveness of a two-stage screening protocol for infants in hospital and community-based settings.
- To evaluate the relative merits and drawbacks of hospital and community-based screening programmes.
- To identify potential risk factors for PCEHL in Nigeria.

Any useful additional data incidental to these principal objectives such as the rates of PCEHL and the characteristics of the study participants will also be highlighted to the extent that they may facilitate a better appreciation of our key findings.

It is intended that appropriate recommendations will be made to the Federal Ministry of Health in Nigeria from the findings of this research to serve as a basis for further actions aimed at implementing the provisions of the Revised National Health Policy and the WHA resolution on hearing impairment prevention. This study however, did not set out to explore the effectiveness of the range of intervention options or communication modalities for children with PCEHL in Nigeria, an objective that is more suited for a longitudinal research well beyond the scope and duration of the present study. Moreover, any initiatives in this direction will have policy implications that extend beyond the remit of the Ministry of Health to include the Ministry of Education and Ministry Youth and Social Development at state and federal levels.

Chapter 2

Literature Review

2 Literature Review

2.1. Auditory System and Language Development

2.1.1. The Concept of Early Childhood Development

The first years of a child's life signify a crucial period when brain development is most susceptible to physiological and experiential influences even as the interactive interplay of both early experience and gene expression has been shown to affect the architecture of the brain and the emergence of life-long capabilities [Shonkoff & Phillips, 2000; Johnson & Blasco, 1997]. Researchers have argued that subsequent robust or fragile foundation for later developmental achievements in children including overall well-being evolve from birth to age 5 years [Shonkoff & Phillips, 2000].

Early childhood development is of great interest to a broad array of disciplines and its significance has been noted by developmental paediatricians, behavioural scientists, neuroscientists, biologists, anthropologists, educators and economists. Four core concepts have emerged to reflect the dynamic and continuous interaction between biology (intrinsic) and experience (extrinsic factors) [Heckman, 2006; Knudsen et al., 2006]:

1. The process of skill development and the architecture of the brain are dependent on the influence of genetics and individual experiences.
2. Hierarchical rules of building later achievements on earlier ones underpin the mastery of skills and the development of their neural pathways.
3. The experiences of the developing child powerfully shape the cognitive, linguistic, social and emotional competencies in early childhood.

4. Evolution of human abilities has been linked to predictable sequence of sensitive periods when the developmental process is most susceptible to environmental influences.

Consequently, every child is expected to develop and acquire functional skills in key interdependent domains of motor (gross and fine); language (receptive and expressive); cognitive; and psychosocial development in early childhood as a basis for later educational and vocational attainment [Johnson & Blasco, 1997; First & Palfrey, 1994].

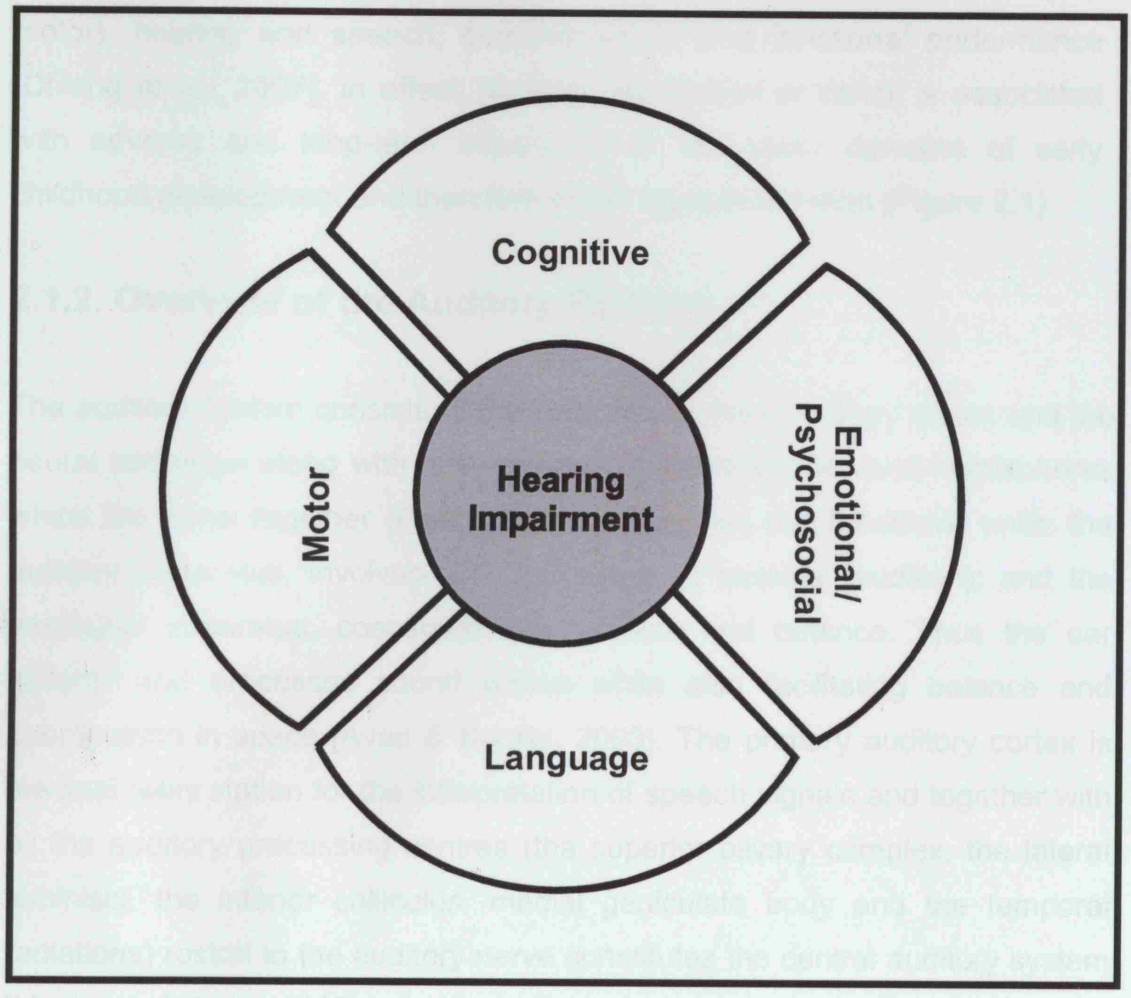


Figure 2.1. Early Childhood Hearing Impairment and Developmental Domains

There is ample evidence that auditory deficit in early childhood affects optimal development of fine motor, receptive and expressive language, cognitive and emotional/psychosocial skills [Chiong et al., 2007; Horn, Pisoni & Miyamoto, 2006; Kennedy et al., 2006; Wake et al., 2004; Moeller, 2000; Yoshinaga–Itano, Sedey, Coulter & Mehl, 1998; Robinshaw, 1995 & 1996; Hindley, 1997; McKellin, 1995; Ramkalawan & Davis, 1992; Ross, 1990; Wiegersma & Van derVelde, 1983; Freeman, Malkin & Hastings, 1975; Vermon, 1969]. For instance, in one study at least 40% of infants detected with hearing loss shortly after birth and followed-up for a period of two years exhibited lower than average scores on Griffiths Mental Development Scale which incorporates locomotor (gross motor), eye-hand coordination (fine motor), hearing and speech, personal social and functional performance [Chiong et al., 2007]. In effect, auditory deprivation or deficit is associated with adverse and long-term effects on all the major domains of early childhood development and therefore merits special attention (Figure 2.1).

2.1.2. Overview of the Auditory Pathway

The auditory system consists of the ears, the primary auditory cortex and the neural pathways along with relay stations in the brainstem and hemispheres which link them together (Figure 2.2). The ear has two functional units: the auditory apparatus, involved with the sense of hearing (audition); and the vestibular apparatus, concerned with posture and balance. Thus the ear collects and processes sound waves while also facilitating balance and coordination in space [Avan & Bonfils, 2003]. The primary auditory cortex is the final relay station for the interpretation of speech signals and together with all the auditory processing centres (the superior olivary complex, the lateral leminisci, the inferior colliculus, medial geniculate body and the temporal radiations) rostral to the auditory nerve constitutes the central auditory system [Musiek & Oxholm, 2003; Luxon, 1981].

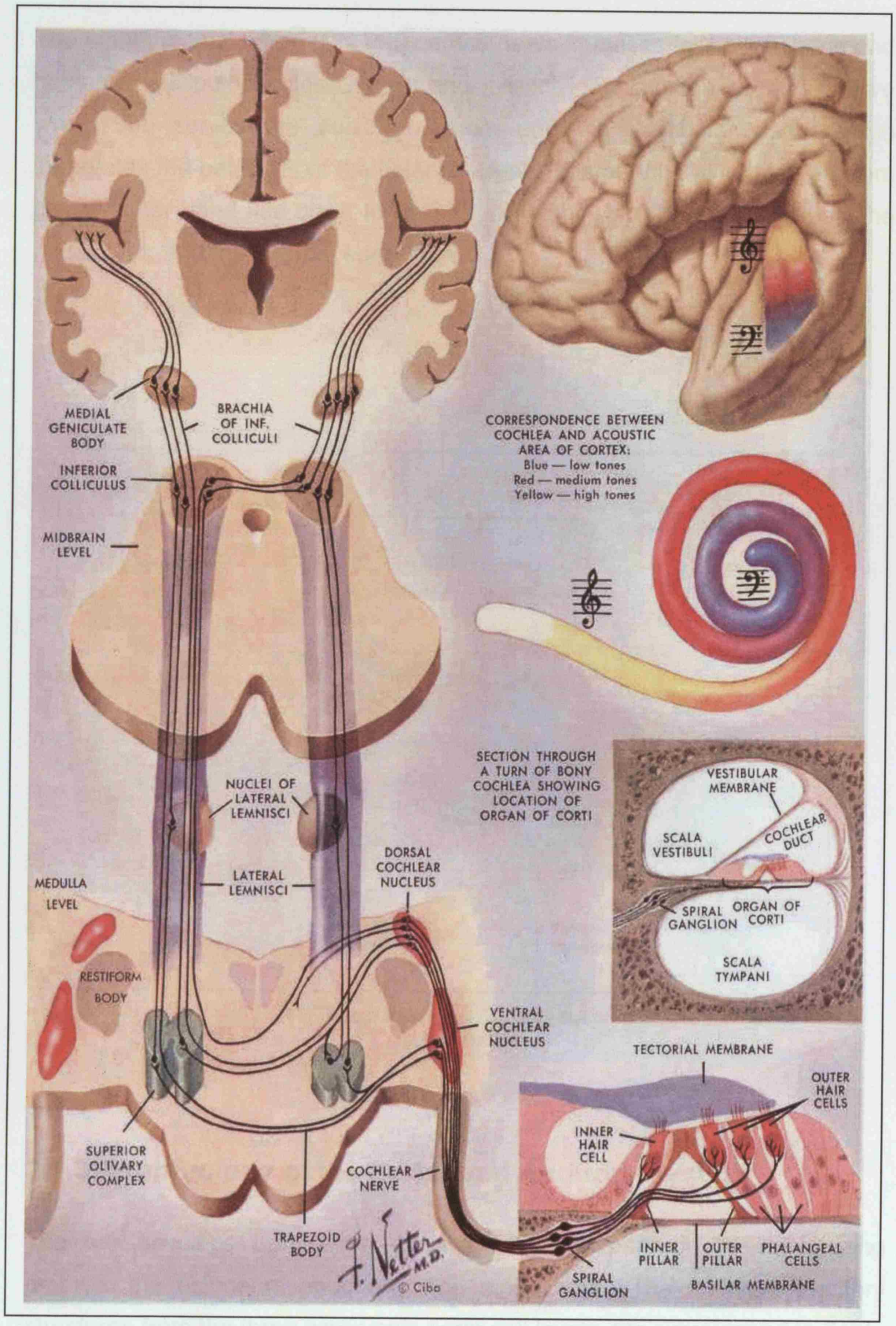


Figure 2.2. Complete Auditory Pathway

[http://pluto.fss.buffallo.edu1.auditorysystem/html]

The eighth cranial nerve (CN VIII) carries nerve impulses for both hearing and balance (vestibulocochlear nerve) and it is divided into the cochlear nerve which sub serves the auditory system and the vestibular nerve which innervates the hair cells of the balance organ. The peripheral auditory system on the other hand lies distal to CN VIII and consists of the external ear, the middle ear; and the internal ear (Figure 2.3).

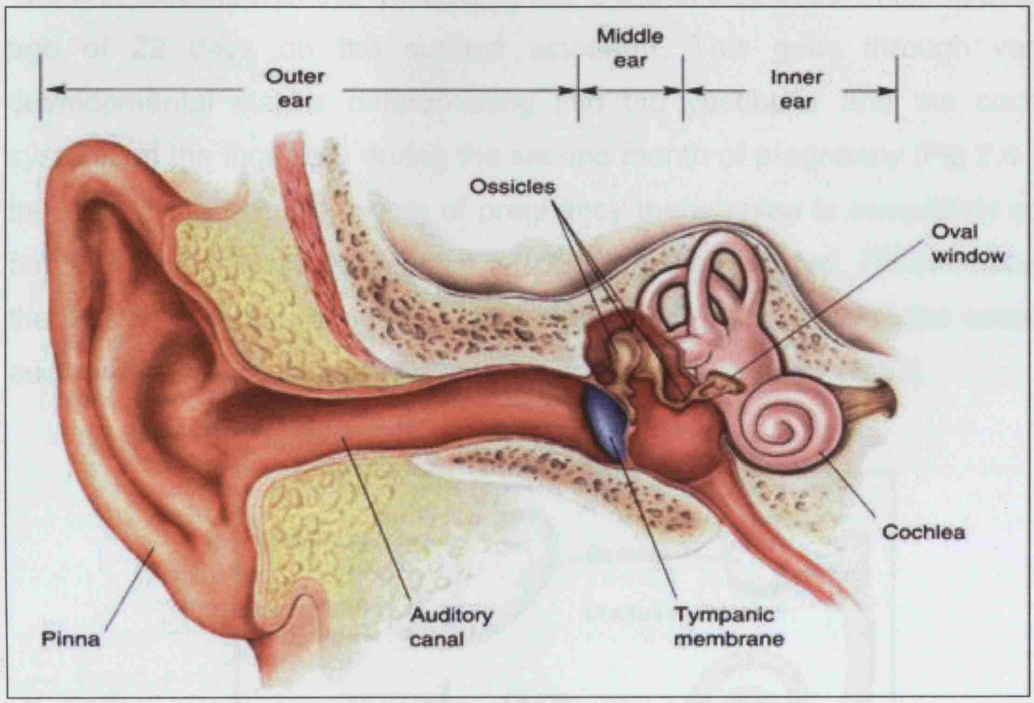


Figure 2.3. Peripheral Auditory System
[www.hoh.ie]

2.1.3. Embryology of the Peripheral Auditory System

The ear develops from three distinctly different parts in the developing embryo: the ectoderm, endoderm and mesoderm. The definitive auditory structures from the three embryonic germ cell layers are summarised in Table 2-1.

Table 2-1. Auditory Structures and the Embryonic Germ Cell Precursors

Embryonic Germ Layers	Definitive Auditory Structures
Ectoderm from 22 days	Cochlea, spiral ganglion, outer layer of tympanic membrane, cartilaginous portion of external auditory canal and pinna
Endoderm from 28 days	Eustachian tube, lower half of the middle ear and medial layer of tympanic membrane
Mesoderm from 49 days	Ossicles (malleus, incus and stapes), mastoid process, temporal bone and middle layer of tympanic membrane.

The first indication of the developing ear is the otic placode which appears at age of 22 days on the surface ectoderm. This goes through various developmental stages differentiating into the vestibular and the cochlear systems of the inner ear during the second month of pregnancy (Fig 2.4.). By the end of the second month of pregnancy the cochlea is completely coiled but its sensory epithelial surface is totally undifferentiated. Differentiation of the hair cells begins at 11-12 weeks of gestation and the adult-like pattern is acquired by the 4th month in utero [Bachmann & Arvedson, 1998].

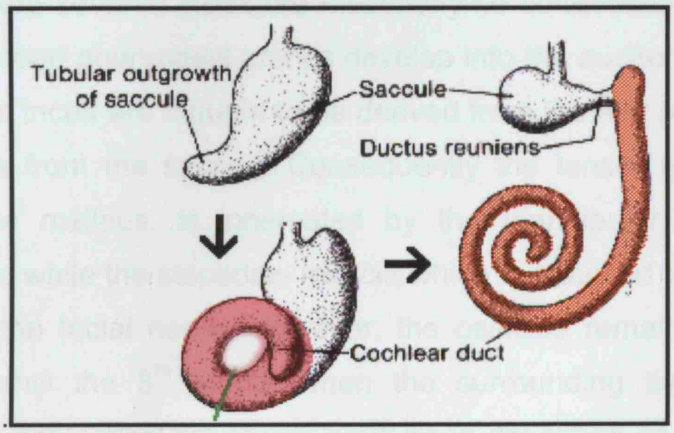


Figure 2.4. Inner Ear Structures
[http://www.med.unc.edu/embryo_images/unit-ear/ear_htms/ear011.htm]

Although the entire length of the cochlea except for the apex is lined with three rows of outer hair cells and a single row of inner hair cells the development and maturation of the outer hair cells lags behind that of the inner hair cells.

The middle ear cavity on the other hand is of endodermal origin. From the first appearance of 'the 1st pharyngeal cleft', at the age of 28 days (Figure 2.5), a rapid development into the middle ear cavity of the Eustachian tube follows.

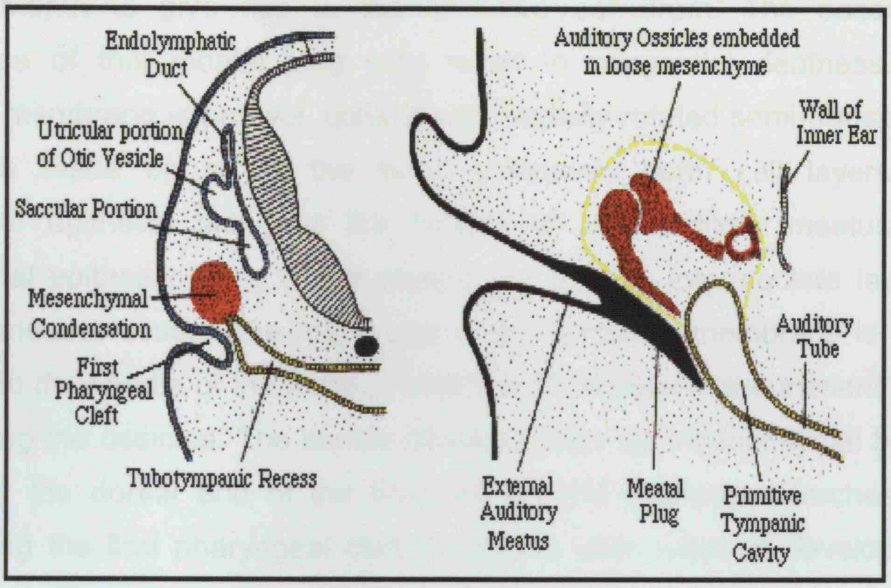


Figure 2.5. Middle Ear Structures
[http://www.med.unc.edu/embryo_images/unit-ear/ear_htms/ear012a.htm]

By the end of the seventh week the mesenchyme condensations caused by the first and second pharyngeal arches develop into the auditory ossicles. The malleus and the incus are thought to be derived from the first pharyngeal arch and the stapes from the second. Consequently the tensor tympani muscle attached to the malleus, is innervated by the mandibular branch of the trigeminal nerve while the stapedius muscle which is attached to the stapes, is innervated by the facial nerve. However, the ossicles remain embedded in mesenchyme until the 8th month when the surrounding tissue dissolves. Although, adult anatomical structures have been described as consequences of both developmental and evolutionary processes from marine to terrestrial life [McLachlan, 2005] the only link to the preceding marine life is the sealed fluid-filled pouch of the inner ear (Figure 2.4).

The external auditory canal develops as a funnel shaped tube from the dorsal portion of the first pharyngeal cleft. A solid epithelial plate known as the meatal plug forms at the bottom of the meatus from the rapid proliferation of epithelial cells at the beginning of the third month. This plug dissolves in the seventh month to give rise to the definitive ear drum. The occasional persistence of this meatal plug may result in congenital deafness. The tympanic membrane is an oval, conical and medially rotated semi-transparent membrane made up of all the three embryonic germ cell layers, the ectodermal epithelial lining at the bottom of the auditory meatus, the endodermal epithelial lining of the middle ear and an intermediate layer of loose connective tissue. The major part of the tympanic membrane is firmly attached to the handle of the malleus after the dissolution of the mesenchyme surrounding the ossicles. The auricle develops from six mesenchymal bulges located at the dorsal end of the first and second pharyngeal arches and surrounding the first pharyngeal cleft during the sixth week of development (Figure 2.6). These hillocks, which first appear at the level of the neck later fuse and are transformed into the definitive auricle or pinna. Further migration brings them to the final position on the side of the head. This complex development which involves migration of the external ear to the definitive position often accounts for frequent anomalies of the external ear.

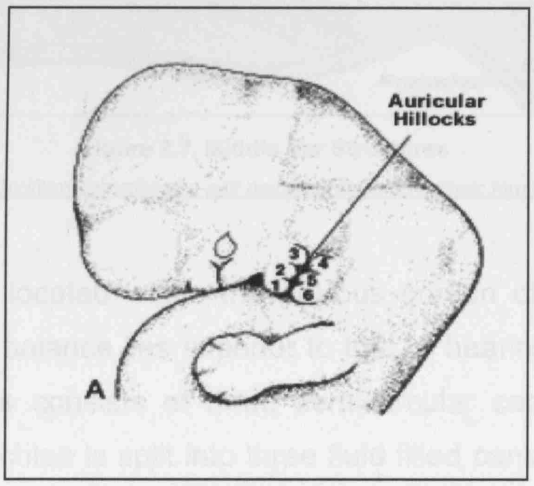


Figure 2.6. External Ear Development

[http://www.med.unc.edu/embryo_images/unit-ear/ear_htms/ear015.htm]

2.1.4. Anatomy of the Peripheral Auditory System

The external ear is made up of the pinna, the external auditory canal and the tympanic membrane (Figure 2.3). The pinna essentially collects sound waves from the environment and channels them into the auditory canal from where they are directed to the tympanic membrane (TM). The TM separates the external and middle ears. Apart from being a resonator, the ear canal also has an indirect effect on auditory function by acting as a buffer against temperature and humidity changes which can alter the elasticity of the tympanic membrane.

The middle ear houses the three ossicles, two muscles (tensor tympani and stapedius muscles), two nerve bundles, the facial nerve and the chorda tympani (a branch of the facial nerve) and the Eustachian tube which acts as a bridge to the pharynx (Figure 2.7).

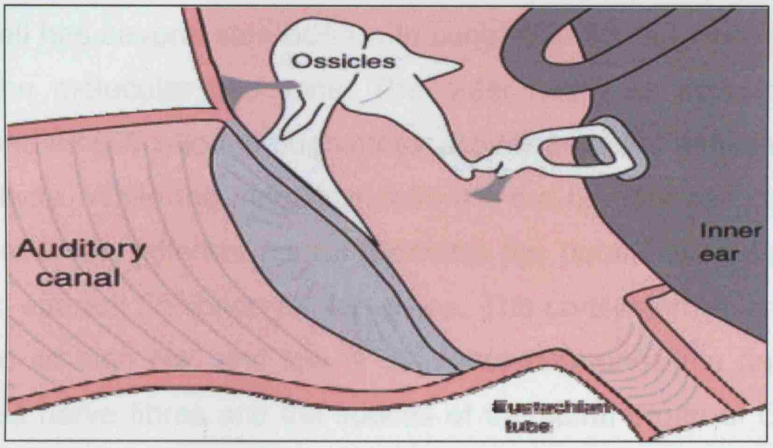


Figure 2.7. Middle Ear Structures

[http://hyperphysics.phy-astr.gsu.edu/hbase/sound/ear.html#c3]

The internal ear is located within the petrous portion of the temporal bone where the organ of balance lies superior to that of hearing (the cochlea). The vestibular apparatus consists of three semi-circular canals and two otolith organs while the cochlea is split into three fluid filled canals. These organs lie in the membranous labyrinth, suspended in perilymph within the bony labyrinth (Figure 2.3).

The cochlea consists of three ducts: the upper scala vestibule, the lower scala tympani and the middle cochlear duct arranged in parallel and coiled in a spiral $2^3/_4$ times around the bony core or modiolus like a snail shell. The scalae vestibule and tympani contain perilymph which is close to the composition of plasma in its high Na^+ and low K^+ contents and communicate with each other at the apex of the cochlea even as they end separately at the oval and round windows respectively. The cochlear duct, located between the two scalae on the other hand, is filled with endolymph (high in K^+ but low in Na^+) secreted by the stria vascularis on the lateral wall. Its roof is the vestibular (Reissner's) membrane bordering the scala vestibule, and its floor, resting on the scala tympani is the basilar membrane. The spiral organ of Corti, the hearing organ, is bounded on top by the reticular (tectorial) membrane and below by the basilar membrane. It includes the sensory receptor or hair cells responsible for hearing.

There are three rows of outer hair cells and one single row of inner hair cells. Each hair cell has several stereocilia with contractile fibres (actin and myosin), acting as the molecular backbone. The outer hair cells provide frequency resolution and amplification through motor activity and also modulate the inner hair cell activity. While the inner hair cells are the primary sensory cells and contain most of the afferent neural terminals the outer hair cells have both afferent and efferent (inhibitory) innervations. The cortilymph which resembles perilymph in its high Na^+ and low K^+ contents surrounds the hair cells, the unmyelinated nerve fibres and the spaces of the spiral organ of Corti (Figure 2.8).

The basilar membrane maintains the ionic composition of the cortilymph by acting as a sieve for ions passing from the perilymph of the scala tympani while the tectorial membrane prevents an exchange of ions between the endolymph and the perilymph. The high Na^+ and low K^+ concentrations of cortilymph maintain a normal physiological ionic environment for the generation and transmission of excitatory impulses.

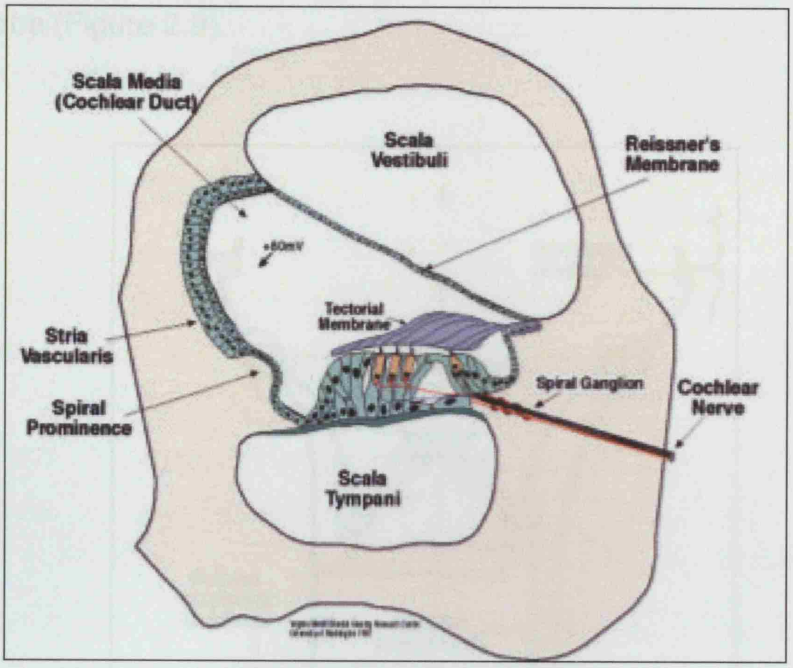


Figure 2.8. Cochlear Hair Cells
[http:// depts.washington.edu]

The central auditory system begins with the cochlea nucleus and traverses the brainstem up to the auditory cortex in the temporal lobe. It consists of the ascending and descending neural fibres with many synaptic stations. The ascending neural pathways from each ear diverge above the level of the cochlear nuclei resulting in bilateral representation in both primary auditory cortices (Figure 2.9). Consequently unilateral lesions above the cochlear nucleus which do not cross the midline do not produce total unilateral hearing loss.

From the dorsal and ventral cochlear nuclei, crossed and uncrossed ascending second order neurons project directly into the lateral leminisci or via inter neurones with relay stations in the nucleus of the trapezoid body or the superior olive. Second and third order neural projections from the lateral leminisci reach the inferior colliculi at the level of the mid brain. The third and fourth order neurones from the inferior colliculus in the mid-brain project to the medial geniculate body in the thalamus from where the fourth order neurones

proceed through auditory radiations to the primary auditory cortex in the temporal lobe (Figure 2.9).

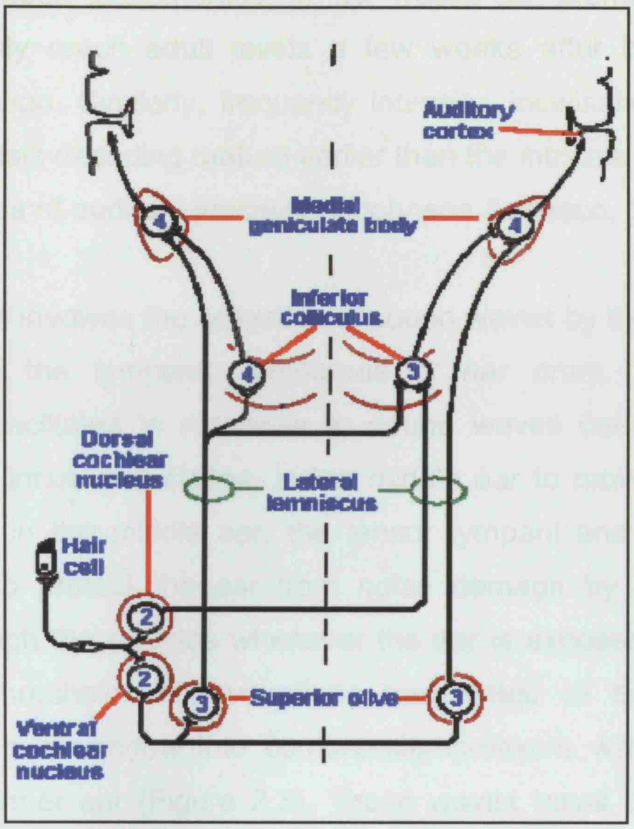


Figure 2.9. Primary Auditory Cortex and The Auditory Pathway
[http://www.conradsimon.org/images/auditory-1.jpg]

2.1.5. Physiology of the Peripheral Auditory System

The hair cell is a common type of receptor cell shared by the auditory and vestibular systems. It is sensitive to movement produced by sound waves, gravity, linear and angular acceleration. A normal adult human ear is capable of hearing very small changes in both the frequency and the duration of sound from approximately 20 to 20,000 cycles per minute (Hz). Humans are thus able to tell a difference as small as 0.1 percent frequency change and can process sounds from the quietest whisper to the blast of a jet engine. By the fifth month of gestation the human cochlea is capable of responding to sound. The simultaneous development of acousticomotor reflexes and

brainstem pathways from the auditory nerve to the auditory cortex around the 28th week of gestation ushers in the onset of synchronized auditory conduction [Hepper & Shahidullah, 1994]. Although adult-like evoked responses have been documented in 35 weeks-old premature infants, latency values only reach adult levels a few weeks after birth following increased myelination. Similarly, frequency intensity, localisation and reflex responses from basic decoding mature earlier than the intricate interpretation of cognitive features of auditory perception [Johnson & Blasco, 1997].

Hearing or audition involves the collection of sound waves by the pinna via the auditory canal to the tympanic membrane or ear drum. The tympanic membrane then oscillates in response to sound waves causing the three ossicles: malleus, incus and stapes, in the middle ear to move sequentially. The two muscles in the middle ear, the tensor tympani and the stapedius muscle contract to protect the ear from noise damage by limiting sound transmission through the ossicles whenever the ear is exposed to sound 80-90 dBA above threshold. The vibrations transmitted to the stapes are converted at the oval window into compression/pressure waves within the perilymph of the inner ear (Figure 2.3). These waves travel up through the scala vestibule to the helicotrema at the apex of the cochlea while concurrently stimulating the endolymph of the cochlear duct. After passing through the helicotrema, the vibrations travel down the perilymph of the scala tympani and in the process stimulate the basilar membrane of the spiral organ of Corti. The vibrations produce a shearing motion of the tectorial membrane which depolarises the stereo cilia of the hair cells. This triggers the release of neurotransmitters at the base of the hair cells (synaptic station) enabling the process of transduction of mechanical energy into electrical signal. The resultant action potentials which are generated are transmitted through the auditory nerve to the primary auditory cortex.

A step down in the amplitude of air-conducted sound waves to match the high energy but low amplitude vibrations through the fluid-filled inner ear (impedance matching) is responsible for the efficient transmission of sound

signals through the peripheral auditory system. The overall efficiency of conducting sound from the air filled spaces by overcoming the impedance mismatch through the fluid filled spaces to the central auditory system is achieved in the following ways:

1. the amplification enhancement of 10 – 20 dB, in the medial part of the auditory canal, close to the TM, especially at 2000 to 5,500 Hz (resonator).
2. buckling deformation of the tympanic membrane which doubles the amplification
3. one and a half times increase in amplification from the lever action of the ossicles
4. sound conduction from the large surface area of the tympanic membrane to the small oval window generating up to 35 times increase in amplification
5. active amplification by the outer hair cells, which also generate otoacoustic emissions in normal ears. This function is now employed for newborn hearing screening.

By far the largest contributor to this process is the difference in the surface areas between the tympanic membrane and the stapes footplate.

The auditory sensory mechanism of the human neonate is fully functional at birth. As early as 19 weeks gestation, hearing in humans can be elicited in response to pure-tone auditory stimuli at 500 Hz. By 27 weeks, the majority of foetuses responded to 250 Hz, while responses to higher frequencies [1000 Hz and 3000 Hz] were documented for all foetuses by 35 weeks gestation [Hepper & Shahidullah, 1994]. Animal experiments have shown that the basal end of the cochlea develops first and responds to low-frequency sounds initially. With increasing maturity and development, responses to low-frequency sounds move rostrally towards the apical region so that the basal region then responds to high frequency sounds [Von-Bekesy, 1960].

Auditory stimulation influences the brain by modifying and customising appropriate circuits involved in processing speech sounds [Knudsen, 2004; Northern & Downs, 2002; Johnson & Blasco, 1997]. The limited period when the effect of experience is particularly strong with respect to development is referred to as the "sensitive period" for that skill. For instance, the capacity to process a language proficiently requires early exposure to the language [Johnson, 2005; Werker & Tees, 2005; Weber-Fox & Neville, 1996; Kuhl 1994]. It has been suggested that stimulation in the sensitive period modifies the architecture of relevant circuits in fundamental ways that cause certain patterns of connectivity to become highly stable and therefore energetically preferred [Knudsen, 2004]. Possible mechanisms of architectural change that underlie plasticity of the sensitive period are [Knudsen, 2004]:

1. Axon elaboration for establishing novel connections instructed by new auditory experiences
2. Synapse elimination of unused synaptic inputs
3. Synapse consolidation of connections that are repeatedly used by highly stable cell adhesion molecules (CAMs) to make them secure and invulnerable to subsequent elimination.

From the knowledge of neural plasticity, it is understood that the compensatory and reorganisation possibilities of the brain are significantly greater in very early childhood than in later life [Kral et al., 2001; Harrison et al., 1991]. It has also been suggested that the individual sensory organs only reach full development if they are intensively stimulated before neural maturation to ensure maximum functional potential [Kral et al., 2001]. These hypotheses have resulted in the emergence of the concept of a "critical phase" for optimal sensory stimulation applicable to the auditory system as well as other structures of the central nervous system, that underpin cognitive development [Eggermont & Ponton, 2003; Ponton, Moore & Eggermont, 1999]. In fact, newborns have been observed to demonstrate selective response to their mothers' voices [Kisilevsky et al., 2003; Mehler, Bertoncini & Barriere, 1978], while infants less than 6 months of age are superior to adults

in their ability to discriminate speech sounds from languages other than their mother tongue. However, the second half of the first year of life reflects a preference for the sounds and pattern of their native language, signifying the beginning of cortical maturation. Linking sound sequences with particular meanings rapidly follows so that a vocabulary of 50 words is attained by age 18 months and 1000 words by 3 years [Kuhl et al., 2006; Rivera-Gaxiola, Silva-Pereyra & Kuhl, 2005; Werker, 2003].

The learning that occurs during the sensitive period lays the foundation for future learning and it is the period during which certain capacities are readily shaped or altered by experience. A sensitive period that provides essential information that permanently alters performance is referred to as a critical period. Critical periods are important because the changes are irreversible. Moreover, the restoration of a typical experience later in life cannot remediate the adverse effects of atypical experience during the critical period. The use of the term critical period therefore refers to a time window with a very specific beginning and end. Sensitive periods are generally longer than critical periods and are acknowledged by more flexibility in the timing of experiences that inform the brain. However, subsequent research has shown that the time window for learning is often flexible and modifiable by experience. Because the period of plasticity is not completely closed in many of the so called critical periods, the term optimal period that shows that plasticity exists outside this window of maximum experiential change, is now preferred [Johnson, 2005; Werker & Tees, 2005]. Thus, it may never be too late to acquire a skill as implied by the concept of a critical period [Johnson, 2005; Werker & Tees, 2005; Weber-Fox & Neville, 1996; Kuhl 1994].

2.1.6. Optimal Period for Speech and Language Development

Although hearing loss has adverse impact on all crucial domains of early childhood development, the effects on speech and language development are more commonly the primary consideration for early hearing detection and intervention programmes particularly in developed countries because of its salutary effects on the other domains.

The effect of hearing loss on language development has been intricately linked to the sensory development of the infantile brain and the nervous system [Hannon, 2003; Stockard-Pope, 2001; Sininger et al., 1999; Ruben & Schwartz, 1999]. Prior to the implementation of universal newborn hearing screening, the period from birth to 5 years was considered as the sensitive phase for the development of language and literacy [Ramey & Ramey, 2006; Carney & Moeller, 1998]. However, in 1986, Markides reported a study on speech intelligibility among four groups of hearing-impaired children fitted with hearing aids at various times and matched for age, gender, age at onset of deafness, degree of hearing loss and the educational establishment attended [Markides, 1986]. Thirty-two children were fitted with hearing aids in their first 6 months of life; 32 children fitted in their second 6 months of life; 38 children fitted in their second year of life and 51 children fitted in their third year of life. Based on teachers rating on a 7-point scale, the speech intelligibility of children fitted with hearing aids in their first 6 months of life was significantly better than the outcomes among the other three groups. Robinshaw [1996 & 1995] in an in-depth study of five infants with severe to profound bilateral sensorineural hearing impairment fitted with hearing aids at 6 months of age and followed up till 21 months of age found that the children acquired vocal communicative and linguistic skills at an age comparable to, or more typical of, their normally hearing peers than infants identified at an average of 27 months following health visitor screening from another study [Tait, 1987]. However, major limitations of the study included the small sample size, short duration of follow-up and lack of a comparative group in the study population of lately-identified children.

Yoshinaga-Itano and her colleagues in Colorado, USA were perhaps the first to provide robust evidence of the superior outcome of early intervention on language development by comparing the receptive and expressive language abilities of 72 deaf or hard-of-hearing children whose hearing losses were identified by 6 months of age with 78 children whose hearing losses were identified after the age of 6 months [Yoshinaga-Itano et al., 1998]. They demonstrated that by age 3 years, early-identified hearing-impaired children

provided with auditory support before 6 months of age, had better language quotients than later-identified children regardless of the degree of hearing loss. Their report was corroborated by subsequent findings that showed significantly better vocabulary, general language abilities (speech intelligibility, phoneme repertoires, syntax), social-emotional development, parental bonding and parental grief resolution in early-identified (by 6 months of age) as opposed to later-identified (beyond 6 months of age) children [Yoshinaga-Itano, 2003; Yoshinaga-Itano, Coulter & Thomson, 2001].

Around this period, Thompson and colleagues undertook a systematic review of the literature to identify the strengths, weaknesses and gaps in the evidence in support of UNHS and whether early detection and intervention by age 6 months improved language outcomes [Thompson et al., 2001].. They reported that there was quality evidence that the current screening technologies were highly accurate and reliable in detecting moderate-to-profound hearing loss and that UNHS increases the chances of early detection and intervention before the age of 6 months significantly. However, they concluded that the effectiveness of UNHS to improve long-term language outcome was uncertain and that the prevailing evidence in support of intervention before 6 months was "inconclusive" because of the lack of high quality randomised controlled trials and methodological flaws associated with the adjustment for cofounders in the widely cited study by Yoshinaga-Itano et al [1998]. Most of the available evidence were also criticised for using convenience samples of subjects, for lacking explicit exclusion criteria and for selecting only children for whom language assessment between ages 2 to 5 years was available. The authors highlighted the need for longitudinal population-based studies involving inception cohorts and a careful reporting of outcomes in all subjects to address these knowledge gaps.

Similarly, a report by the U.S. Preventive Services Task Force [2002], which was an update of an earlier review published in 1999, also concluded that evidence to determine whether earlier treatment by 6 months of age resulting from screening leads to clinically important improvement in speech and

language skills at age 3 years or beyond was inconclusive because of the design limitations in the studies in existence at that point in time. The available evidence was considered insufficient to recommend for or against routine screening of all newborns before hospital discharge. The report further acknowledged that earlier identification and intervention may improve the quality of life for the hearing-impaired infant and the family during the first year of life and also prevent regret by the family over delayed diagnosis of hearing loss but noted the lack of data that substantiate these benefits.

The reservations expressed in these two critical reviews were not entirely shared by some researchers and relevant professional groups that had previously endorsed UNHS such as the JCIH, National Institutes of Health and Centers for Disease Control and Prevention. For example, Yoshinaga-Itano [2004] refuted the claim that their widely cited study published in 1998 was based on a convenience sample and also argued that it was practically impossible to adjust for all confounding factors as suggested in these reviews. The author emphasised the need to distinguish between the many goals served by UNHS such as: to facilitate optimal developmental outcomes for the life of a hearing-impaired child, ensure early identification of PCEHL, achieve early intervention initiation for the child and family or facilitate early access to communication and language development. For most of these objectives, UNHS begins as part of the medical/health system but outcome is largely dependent upon the educational system which often is beyond the control of health professionals. Optimal developmental outcomes particularly in speech and language skills were therefore not only dependent on the age of detection of PCEHL but also on the age and quality of intervention services provided [Yoshinaga-Itano, 2004].

Subsequently, several longitudinal studies have attempted to establish the value of UNHS on long-term language development beyond age 3 years. For instance in 2000, Moeller in her study of 112 deaf and hard-of-hearing children assessed at age 5 years corroborated the view of superior long-term language outcome with early intervention by reporting significantly better vocabulary and

reasoning skills that approximate those of their hearing peers for children enrolled earlier than 11 months and for whom parental involvement was high [Moeller, 2000]. This achievement was attributable to the early initiation of intervention which kept language delay from increasing rather than in later-identified babies where an attempt has to be made to close established delays that would require the children to make greater language gains than typically normal hearing children. Consequently, the time of initiating intervention emerged as an important predictor for successful language outcomes [Yoshinaga-Itano, 2003; Moeller 2000].

In contrast, Wake and her colleagues [2005] argued that the degree of hearing loss rather than the timing of intervention was the decisive predictor of a successful outcome but failed to demonstrate this in an heterogeneous population of 89, 7-8 year old hearing impaired children, a third (29 children) of which were only identified by twelve months of age. However, a recent study from the UK corroborates earlier reports highlighting the timing of intervention as the critical predictor of a successful outcome [Kennedy et al., 2006]. Kennedy and colleagues demonstrated better receptive language outcomes at a mean age of 7.9 years in those who had intervention by 9 months but not with expressive language outcomes from their initial cohorts in the highly rated Wessex controlled trial study [1998]. A related UK study reported by Watkin et al [2007] found significant positive correlations between active family participation and language/speech intelligibility ratings even in children with severe or profound hearing loss who were diagnosed after 9 months of age, thus suggesting that family participation in the intervention programme may significantly compensate for the deleterious effects of late intervention. This finding was derived from a longitudinal study of two separate birth cohorts from the first UNHS programme in the UK at Whipps Cross Hospital [Watkin, Baldwin & McEnery, 1991] and the Wessex Trial Group [1998].

Apart from the foregoing studies which focused on spoken language development, studies among deaf children have shown that the early

introduction of sign language from birth was also more beneficial for early mother-child communication with favourable impact on cognitive and psychosocial development [Schick, Marschark & Spencer, 2006; Meadow-Orlans et al., 2004; Hindley & Parkes, 1999].

These reports altogether would suggest that the first year of life was crucial to favourable language (and related developmental) outcomes in children with hearing loss. The evidence provided by these well-conducted observational/longitudinal studies are considered acceptable in the absence of "ideal" randomised controlled trials which are in fact constrained by ethical reasons for PCEHL particularly in developing countries [Potts et al., 2006; Puig, Municio & Meda, 2005; Victora, Habicht & Bryce, 2004a; Black, 1996; Sackett et al., 1996].

2.2. Evolution of Universal Infant Hearing Screening

2.2.1. Classification of Childhood Hearing Loss

Congenital and early-onset hearing loss can be classified according to the type of loss, time of onset and the causality. Hearing loss affecting the external or middle ear is conductive and usually transient. However, conductive hearing loss associated with structural defects or chronic otitis media may be permanent. Hearing loss is sensorineural and permanent when the cochlea or the eighth nerve as far as the brain is involved. Mixed hearing loss results from involvement of both the conductive and sensorineural components. Sensorineural hearing loss is the commonest congenital abnormality in newborns [Finitzo & Crumley, 1999; Mehl & Thomson, 1998].

From a recent systematic review of the literature on the aetiology of bilateral sensorineural hearing loss in children, the "unknown" group may be up to 38% in developed countries [Mozaria, Westerberg & Kozak, 2004] and could be nearer 60% in developing countries [Sellars & Brighton, 1983]. In fact, two reports derived from population-based studies in Australia and USA

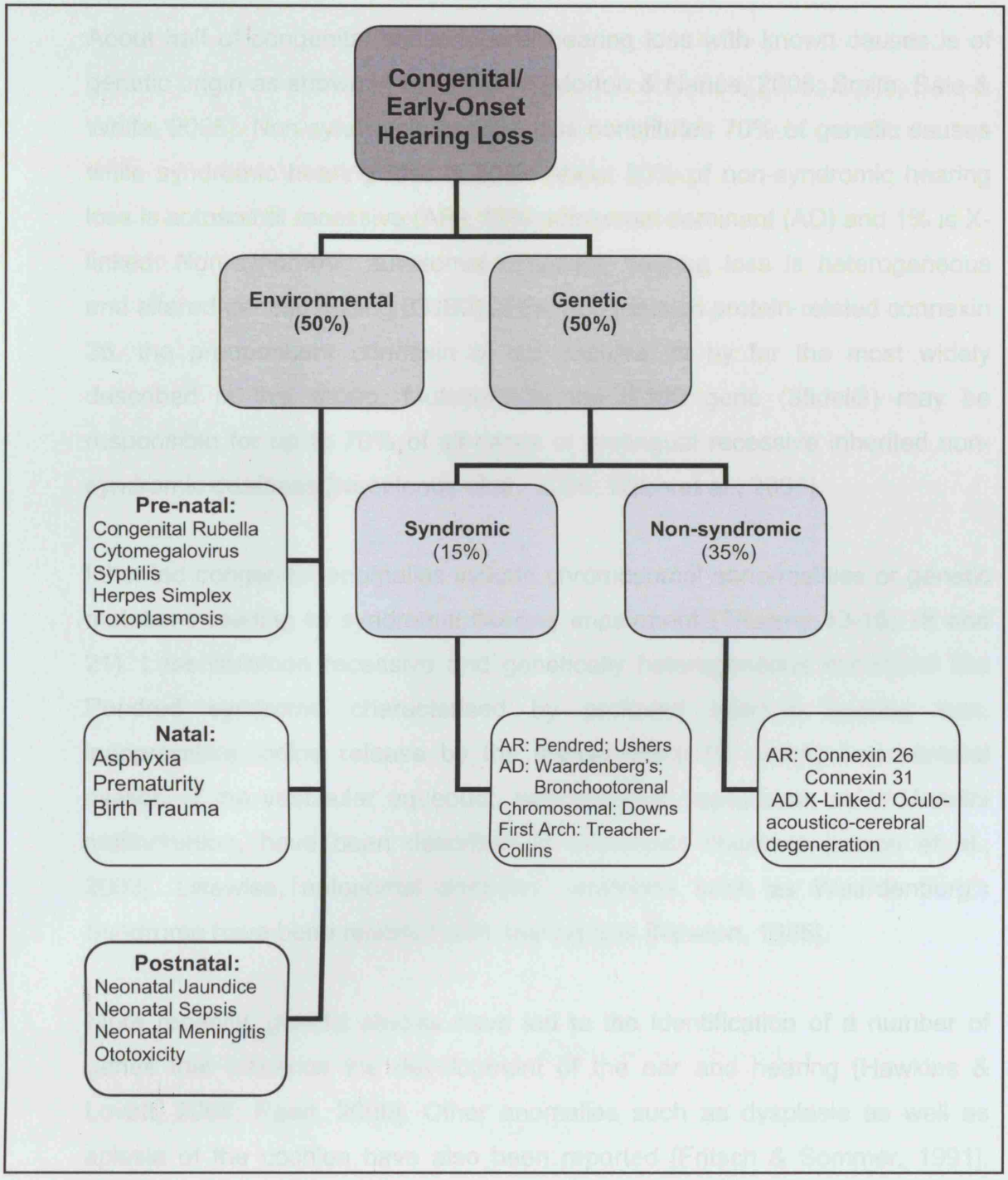


Figure 2.10. Aetiological Contributions to PCEHL

[Adapted from: Smith, Bale & White, 2005]

documented unknown aetiology of 57% and 78% respectively [Russ et al., 2003; van Naarden, Decoufle & Caldwell, 1999].

About half of congenital sensorineural hearing loss with known causes is of genetic origin as shown in Figure 2.10 [Morton & Nance, 2006; Smith, Bale & White, 2005]. Non-syndromic hearing loss constitutes 70% of genetic causes while syndromic hearing loss is 30%. About 80% of non-syndromic hearing loss is autosomal recessive (AR), 19% autosomal dominant (AD) and 1% is X-linked. Non-syndromic autosomal recessive hearing loss is heterogeneous and altered genetic coding (GJB2) of the gap-junction protein-related connexin 26, the predominant connexin of the cochlea, is by far the most widely described in this group. Mutations in the GJB2 gene (35delG) may be responsible for up to 70% of all cases of prelingual recessive inherited non-syndromic deafness [Neocleous et al., 2006; Wilch et al., 2006].

Inherited congenital anomalies include chromosomal abnormalities or genetic mutations leading to syndromal hearing impairment (Trisomy 13-15, 18 and 21). Less common recessive and genetically heterogeneous conditions like Pendred syndrome characterised by profound bilateral hearing loss, inappropriate iodine release by the thyroid gland (or goitre) and bilateral dilation of the vestibular aqueduct with cochlear hypoplasia, as in Mondini malformation, have been described in developed countries [Luxon et al., 2003]. Likewise, autosomal dominant conditions such as Waardenburg's Syndrome have been reported with hearing loss [Newton, 1985].

More recently, genetic studies have led to the identification of a number of genes that influence the development of the ear and hearing [Hawkins & Lovett, 2004; Read, 2000]. Other anomalies such as dysplasia as well as aplasia of the cochlea have also been reported [Fritsch & Sommer, 1991]. Although minor structural abnormalities in the complex development of the pinna, are often not reported with associated hearing impairment, major changes tend to signify serious auditory and other internal anomalies such as the first arch syndrome/mandibular dysostosis (Treacher Collins Syndrome) and other chromosomal anomalies.

Acquired causes of permanent hearing loss resulting from marked structural and functional damage of the auditory system include intrauterine infections with toxoplasmosis, cytomegalovirus, rubella, herpes and syphilis [Newton & Vallely, 2006; Northern & Downs, 2002]. Congenital rubella syndrome presenting with a classical triad of sensorineural hearing loss, cardiac anomalies and cataract was common in the developed countries prior to the advent of rubella vaccination for girls in 1969 in the USA. However, the introduction of the rubella vaccine in 1969 to the developed world led to a substantial reduction in the incidence of congenital rubella internationally but this is not the case in the developing countries where rubella vaccination is currently excluded from many national immunisation programmes [UNICEF, 2006].

Other acquired causes of significant permanent hearing impairment include adverse perinatal conditions such as severe birth asphyxia from prolonged obstructed labour. Teratogens such as thalidomide for instance, ototoxicity e.g. aminoglycosides and metabolic conditions such as kernicterus may also affect the developing ear. These perinatal events are significant causes of progressive and delayed-onset permanent sensorineural hearing loss in developing countries [Olusanya & Okolo, 2006; Newton, 2001]. Other acquired causes of hearing impairment include maternal conditions such as, diabetes and pregnancy-induced hypertension.

2.2.2. Case Definition for Early Hearing Detection

The term "hearing impairment" is used as a generic term to describe the loss of hearing sensitivity in one or both ears and it is therefore used interchangeably with "hearing loss". Hearing impairment in early childhood is generally considered as significant when its duration and degree is capable of causing auditory deprivation that would interfere with normal speech and language development. By duration, hearing loss must be persisting or permanent to have a significant impact on speech and language development. This would include sensorineural hearing loss and permanent (rather than transient) conductive hearing loss. Although hearing thresholds of 16 dB HL –

30 dB HL may not be normal [Northern & Downs, 2002], some researchers do not associate this range with major adverse impact on speech and language development [Norton et al., 2000a; American Academy of Pediatrics (AAP), 1995].

While there is ample evidence that moderate-to-profound bilateral permanent hearing loss (≥40 dB HL) in early childhood significantly impedes speech, language and cognitive development, available evidence also suggest that even children with mild or unilateral permanent hearing loss may experience difficulties with speech, language, educational and psycho-social development [Downs, 2007; Teasdale & Sorensen, 2007; Ruscetta, Arjmand & Pratt, 2005; Cho-Lieu, 2004; Welsh et al., 2004; Bess, Dodd-Murphy & Parker, 1998; Davis et al., 1986; Culbertson & Gilbert, 1986; Blair, Peterson & Viehweg, 1985; Keller & Bundy, 1980]. For instance, in a large cohort of young men attaining the age of 18 years in Denmark due to be drafted into military service, 20% had mild hearing loss and the odds of not attending a senior college was 1.4 times significantly greater than those with normal hearing [Teasdale & Sorensen, 2007]. In addition, it has been argued that auditory deprivation caused by variable hearing impairment during recurring episodes of otitis media may account for the often reported developmental impairment [Paradise, 1998].

However, based on the established principles of health screening, current limitations of available screening technologies and the prospects of effective intervention, the JCIH considers permanent bilateral or unilateral sensory or conductive hearing loss averaging 30 to 40 dB HL or more in the frequencies important for speech recognition (approximately 500 to 4000 Hz) as the target for early hearing detection programmes [Strong et al., 2005; JCIH, 2000; Norton et al., 2000a; Wilson & Jungner, 1968].

In contrast, the target of most early hearing detection programmes in the UK is permanent bilateral hearing loss ≥40 dB HL [Kennedy et al., 2006; Uus & Bamford, 2006; Davis et al., 1997]. Davis et al (1997) had argued that mild or

unilateral hearing loss did not constitute serious public health hazard although their effects on speech perception in noisy or less-than-optimal listening environments and on educational achievement could not be altogether ignored [Hicks & Tharpe, 2002; Bess, Dodd-Murphy & Parker, 1998]. Similarly, Wake et al [2006] more recently found that in an Australian population of school children slight/mild hearing loss did not have adverse effects on language, reading, behaviour and health-related quality of life except phonologic short term memory. However, from an ethical standpoint, it is probably difficult to deny parents the knowledge of any degree of "hidden" impairment in their babies that is detectable by available technologies. For instance based on the current International Classification of Functioning for various health conditions [WHO, 2001], slight/mild hearing loss is associated with some activity limitation and participation restriction as shown in Table 2-2 [Olusanya & Newton, 2007, Clark, 1981].

Bess and colleagues (1998) have in fact argued that the use of the term "mild" or "minimal" may misrepresent or trivialise the potential negative effects of these categories of hearing loss [Bess, Dodd-Murphy & Parker, 1998]. It is not uncommon for mild hearing loss to signal the onset of a progressive and severe hearing loss that requires surveillance and timely intervention [Iwasaki et al., 2007; Fowler et al., 1999; Williamson et al., 1992]. For instance, Downs [2007] reported a study in Colorado in which among 30 babies identified with unilateral hearing loss at birth, two (7%) had progressed to bilateral hearing loss within the first year of life and two (7%) were later diagnosed with mild bilateral hearing loss that could have been present at birth but was missed as a result of the limitation of the screening threshold of 30 dB HL.

WHO defines "disabling hearing impairment" in children under the age of 15 years as a permanent unaided hearing threshold level in the better ear of 31 dBHL or more using pure-tone average over octave frequency levels 0.5, 1.0, 2.0 and 4.0 kHz [WHO 2006a]. However, this classification excludes children with unilateral hearing loss of any degree as well as those with mild hearing loss and does not take into account the current WHO's International

Table 2-2. Common effects of untreated childhood hearing impairment*

Average Hearing Level and Degree of Hearing Impairment*	Receptive Language	Expressive Language	Activity Limitation / Participation Restriction
0 -15 dB HL: *Normal Hearing*	Detects all speech signals	Normal range	None
16 - 25 dB HL: *Slight*	Misses up to 10% of speech sounds (e.g. unvoiced consonants) especially in difficult listening situations	Mild dysfunction in language learning	- Inappropriate response to sound - Learning difficulties - Poor social interaction
26 - 40 dB HL: *Mild*	Misses 25 to 40% of speech especially in difficult listening situations	Mild language retardation and speech problems	- Inattention - Learning difficulties - Behaviour problems
41 - 55 dB HL: *Moderate*	Misses 50 to 75% of speech	Moderate language retardation and poor speech intelligibility	- Learning dysfunction - Significant social problems
56 - 70 dB HL: *Moderately Severe*	Misses 75 to 100% of speech	Severe language retardation and speech problems	- Severe learning dysfunction - Stigmatisation and possible social isolation
71 - 90 dB HL: *Severe*	Misses up to 100% of speech at conversational level	Severe speech problems and language retardation	- Severe learning dysfunction - Stigmatisation and significant social isolation
> 90 dB HL: *Profound*	Misses all loud speech sounds except vibrations.	Visual cues essential for communication	- Complete social isolation

* Source: Olusanya & Newton, 2007

Classification of Functioning [WHO, 2001]. For a country like Nigeria where preschool and school hearing screening are non-existent, the knowledge of a minimal PCEHL at birth that may potentially affect future school performance adversely should help to place the affected children under close parental surveillance.

Such an intervention may also help in identifying cases where mild or unilateral hearing loss deteriorates significantly after the initial detection. Consequently, the case definition for this research project includes mild and unilateral PCEHL in line with JCIH recommendations.

2.2.3. Public Health Perspective of Screening for PCEHL

It has been suggested that at least 50% of the burden of hearing loss in all age groups can be prevented [WHO, 2006a; Smith, 2003; Alberti, 1996]. Prevention in a generic sense consists of three levels of disease control and management: primary, secondary and tertiary.

Primary prevention is concerned with the prevention of the occurrence of any condition that may lead to PCEHL and include such activities as immunisation, the avoidance or rational use of ototoxic drugs, improved obstetric care, personal hygiene and living conditions. For primary prevention to be effective it would entail the accurate knowledge of the causes and the associated risk factors for PCEHL in a given setting [Morzaria et al., 2004]. However, the possible causes of PCEHL in a significant proportion of studies among children with hearing loss in the developing world are unknown and this forestalls primary prevention as an adequate solution [Derekoy, 2000; Minja, 1998; Elango, Chand & Purohit, 1992; Gray, 1989; Sellars & Brighton, 1983; Holborow, Martinson & Anger, 1982].

Moreover, vaccinations against notable causes of PCEHL such as meningitis, mumps and rubella are rare in many developing countries. The standard of maternal and child health care in many of these countries is poor and unlikely to reach levels comparable to those of developed countries in the foreseeable future. Secondary prevention involves actions to prevent PCEHL from becoming a disability through early detection by infant hearing screening and provision of appropriate/timely intervention. Secondary prevention is therefore imperative as a complementary strategy in view of the limitations of primary intervention and current possibilities for early detection.

Screening is the systematic application of a test or enquiry to identify individuals at sufficient risk of a specific disorder to benefit from further investigation or direct preventive action, among people who have not sought medical attention because of symptoms of that disorder [Strong et al., 2005]. The process of screening should identify infants with PCEHL for whom further action is warranted (test-positives) and infants without PCEHL for whom no further action is warranted (test-negatives). It is highly unlikely that any hearing screening test can accurately distinguish all infants with PCEHL from those without due to the inherent differences in biomedical investigation and test algorithms. Consequently, a hearing screening test usually results in four main outcomes:

A. Infants with PCEHL accurately identified (True-Positives)
B. Infants without PCEHL accurately identified (True-Negatives)
C. Infants with PCEHL not accurately identified and classified as having normal hearing (False-Negatives)
D. Infants without PCEHL not accurately identified and classified as having abnormal hearing (False-Positives)

The performance of an infant hearing screening test is therefore evaluated on the basis of the following parameters:

- Sensitivity – probability of a positive test in children with hearing loss or the percentage of children with hearing loss correctly detected.
- Specificity – probability of a negative test in children without hearing loss or the percentage of children without hearing loss correctly detected as having normal hearing.
- False Positive Rate (FPR) – probability of a child without hearing loss testing positive or the percentage of children without hearing loss who had positive test results.

- Positive Predictive Value (PPV) – probability of a child having hearing loss when the test is positive or the percentage of those with positive test results who actually have hearing loss.
- Positive Likelihood Ratio (PLR) – the likelihood that a positive test result will be found in patients with hearing loss compared to patients without hearing loss. In effect, PLR tells us how much more likely a positive test is to be found in patients with hearing loss as opposed to patients without hearing loss.
- Negative Likelihood Ratio (NLR) – the likelihood that a negative test result will be found in patients without hearing loss compared to patients with hearing loss. In effect, NLR tells us how much more likely a negative test is to be found in patients without hearing loss as opposed to patients with hearing loss.

Whilst screening has the potential to improve quality of life through early diagnosis, the process still has some limitations [National Screening Committee, U.K., 2000]. Screening can reduce the risk of developing a condition or its complications but it cannot offer a guarantee of protection. In any screening programme, there is an irreducible minimum of false positive results (wrongly reported as having the condition) and false negative results (wrongly reported as not having the condition). In practice, an ideal hearing screening test would be simple to apply, safe, reliable and valid. It is reliable if it provides consistent results and valid if it detects the majority of children with hearing loss (high sensitivity); does not pick most children without hearing loss as failing the test (high specificity) or the percentage of children without hearing loss among those with positive test results is very low (low FPR); and if the percentage of those with hearing loss among those with positive test results is high (high PPV).

Tertiary prevention principally entails the rehabilitation of persons with disabilities. Rehabilitation has been defined as "the use of all means aimed at reducing the impact of disabling and handicapping conditions and enabling

people with disabilities to achieve optimal social integration" [Gutenbrunner, Ward & Chamberlain, 2006]. Tertiary prevention for PCEHL therefore seeks to prevent the disability posed by restricted hearing sensitivity from becoming a disability that limits participation in society at large [WHO, 2001]. It embraces the provision of hearing aids or cochlear implants and associated services, enrolment in family-oriented support services, special education and access to social integration [WHO, 2006a]. Social integration or participation requires a matching of the social and physical environment to the needs of children with PCEHL, so as to remove societal barriers to participation, be they environmental, social, educational or vocational.

2.2.4. Historical Perspectives to Early Hearing Detection

The history of UNHS is often linked to the pioneering work of Sir Alexander and Lady Ethel Constance Ewing of Great Britain in 1944 when they highlighted the need to develop methods of testing hearing in young children in the first year of life [Ewing & Ewing, 1944]. They were among the earliest to investigate systematically infant behavioural responses (sound recognition) to auditory stimuli using sounds from toys, noisemakers, crumpled paper and the human voice. They also documented unconditioned response by eye-shifts and head turns in the direction of sound (sound localisation). In 1946, Froeschels and Beebe investigated hearing loss in newborns and reported that infants responded to whistles but not to tuning forks, which were used for older children. Hardy and co-workers in 1959 evaluated auditory response of newborns to doorbell, tonette and "clacker", a custom-made device that produced a broad-spectrum signal. They extended their work to investigate the aetiology of deafness, which was ultimately developed into the High-Risk Register for neonatal hearing loss. Between 1964 and 1969, Downs and her colleagues applied behavioural screening to neonates using a device that generated a narrow band noise of 90 dB to elicit response but was only able to detect infants with severe-to-profound hearing loss and thereby resulted in high false-positives and high false-negatives [Downs & Hemenway, 1969; Downs & Sterritt, 1964].

Further interest in neonatal and infant hearing screening led to the formation of the US Joint Committee on Infant Hearing (JCIH) in 1969 to improve and coordinate the efforts for the early detection of congenital hearing loss. As a result of the limitations with the available behavioural screening techniques and methodologies the JCIH in 1972 recommended the screening of only high-risk babies, based on a set of five risk factors, which were by 1990 and 1994 expanded to ten [American Academy of Pediatrics (AAP), 1995] but more commonly grouped into the following five categories [JCIH, 2000]:

1. *An illness or condition requiring NICU admission for 2 days or more.*
2. *Stigmata or other findings associated with a syndrome known to include sensorineural and/or conductive hearing loss.*
3. *Family history of sensorineural hearing loss.*
4. *Cranio-facial anomalies, including those with morphologic abnormalities of the pinna and ear canal.*
5. *In-utero infections such as rubella, cytomegalovirus, syphilis, toxoplasmosis, bacterial meningitis and herpes.*

Meanwhile, in 1974, Simmons and Russ [1974], in order to address the observer bias that was the principal flaw with the behavioural methods, developed the Crib-o-gram, which was an automated screening device. It used a motion detector under the mattress in the baby's crib to measure the baby's response to a narrow band signal at around 75 dBA. However, this device was found to be unreliable in neonatal intensive care unit and unsuitable for short-stay well babies in hospitals.

In 1979, Bennett in the UK developed the Auditory Response Cradle (ARC) which utilized a technique similar to the Crib-o-gram. It added the detection of a physiological response into behavioural response evaluation of babies. Unfortunately, the device was found to be unsuitable for mass screening because of its high cost, large size and relatively high intensity of the test signal. However, the technique paved the way for the introduction of physiological tests in newborn hearing screening. Fortuitously, two physiologic

measures - Auditory Brainstem Response (ABR) and Otoacoustic Emissions (OAE) - emerged around this period for screening newborns.

The ABR was first discovered between late 1960s and early 1970s by some Israeli and American investigators, but its application for newborn hearing screening was not described until nearly a decade after by Schulman-Galambos and Galambos [1979]. The ABR is an electro-physiological measure of the function of the auditory pathway from the eighth cranial nerve through the brainstem as shown in Figure 2.9. The major advantage of this test which is the electrical recording from three surface scalp electrodes to auditory stimuli is the fact that it is not state-dependent as recordings can be obtained when babies are sleeping or sedated. In addition, the response is significantly correlated with the degree of hearing loss. In general, the click-evoked threshold predicts behavioural audiometric threshold in the 1,000 to 4,000 Hz range within 10 to 15 dB HL. It was therefore found valuable as a confirmatory test in newborn screening [Hyde, Riko & Malizia, 1990; Kileny & Magathan, 1987]. Automated ABR version (AABR) soon followed and was designed as a screening technique to produce simply a 'pass' or 'fail' result. Most AABR units are portable, simple to use and do not require audiological expertise. A typical ABR wave-form is shown in Figure 2.11.

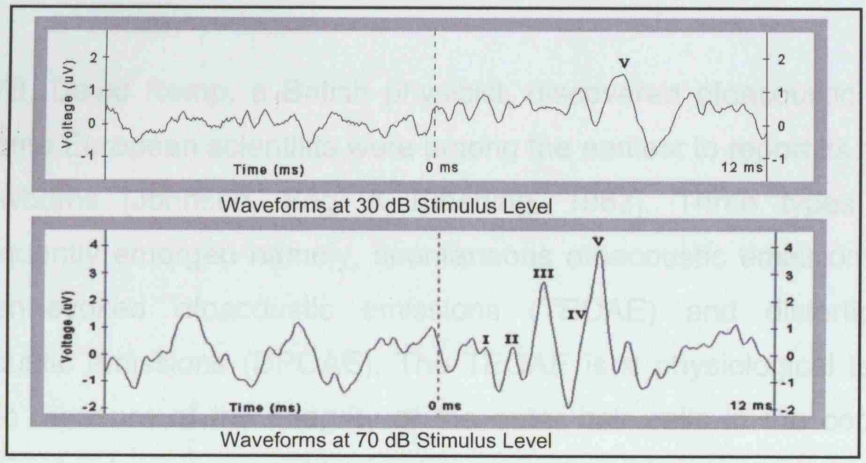


Figure 2.11. Typical ABR Waveforms

- The ABR wave I response is believed to originate from afferent activity of the cranial nerve (CN) VIII fibres (first-order neurons) as they leave the cochlea [Picton et al., 1974].

- ABR wave II is generated by the proximal VIII nerve as it enters the brain stem through the internal auditory canal [Moller, Jannetta & Sekhar, 1988; Picton et al., 1974].

- The ABR wave III arises from second-order neuron activity. The cochlear nucleus, the trapezoid body and the superior olivary complex have been suggested as possible sites of origin for wave III [Picton et al., 1974].

- The ABR wave IV, which often shares the same peak with wave V, is thought to arise from pontine third-order neurons mostly located in the superior olivary complex, but additional contributions may come from the cochlear nucleus and nucleus of lateral leminiscus [Pratt, 2003].

- The ABR wave V is believed to originate from the vicinity of the inferior colliculus and it is the component analyzed most often in clinical applications. Thalamic (medial geniculate body) origin is suggested for generation of waves VI and VII, but the actual site of generation is uncertain [Picton et al., 1974].

In 1978, David Kemp, a British physicist, discovered otoacoustic emissions and some European scientists were among the earliest to report its application to newborns [Johnsen, Bagi & Elberling, 1983]. Three types of OAEs subsequently emerged namely, spontaneous otoacoustic emissions (SOAE), transient-evoked otoacoustic emissions (TEOAE) and distortion-product otoacoustic emissions (DPOAE). The TEOAE is a physiological test for the specific measure of the integrity of the outer hair cells in the cochlea. The TEOAE, also known as cochlear echoes, are low intensity sounds originating from the active amplification of the outer hair cells and can be elicited in

response to clicks presented to the ear through a light weight probe that houses both a transducer and microphone/receiver (Figures 2.12).

Figure 2.12. Typical Recording of a Response from a Stimulus
[Insert: Handheld TEOAE Screener-Echocheck- by Otodynamics UK].

The emissions are then matched through advanced digital processing technology with a normal template before giving a 'pass' or 'refer' result. Most TEOAE screeners simply produce a 'pass' or 'fail' result while the non-portable models usually display the waveforms as shown in Figure 2.12.

The test is relatively quick, non-invasive and does not require sleep or sedation, which makes it readily tolerable by babies and acceptable to parents. The TEOAE screener is lightweight and easy to use. The recording often takes seconds and can be administered without audiological expertise. The sensitivity is greater than 90% and specificity may be up to 99% based on two stage screening [Johnson et al., 2005; Lin et al., 2005; Hall 3rd, Smith & Popelka, 2004; Watkin, 2003; Vohr et al., 2001; Gravel et al., 2000; Kennedy et al., 2000]. One disadvantage with this test in the newborn is that it is sensitive to mild conductive hearing loss resulting from debris associated with

vernix caseosa and amniotic fluid in the external ear canal [Doyle et al., 2000; Maxon et al., 1997; McNellis & Klein, 1997]. While the technique is sensitive to peripheral hearing impairment it will not detect auditory neuropathy. In contrast, the DPOAEs are generated by two continuous pure tones introduced to the ear simultaneously. Because DPOAE are evoked by frequency-specific signals, it is possible to use the response to predict frequency-specific hearing sensitivity across the range 500 to 8,000 Hz. However, this advantage is not critical for screening infants and young children.

While the AABR tests the peripheral auditory pathway from the ear through the brainstem, TEOAE tests the peripheral hearing pathway of the ear through the middle ear to the internal ear (Figure 2.13).

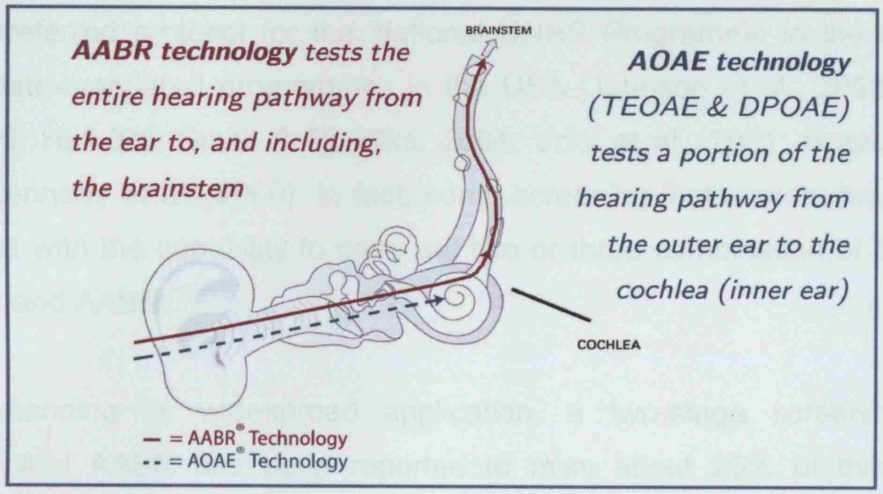


Figure 2.13. Pathways for Hearing Screening Tests
[http://www.natus.com/products/hearing_screen/echo.html]

Some babies with hearing loss may have normal outer hair cell function but a dysfunction in neural transmission through the spiral ganglion cells, their processes and the 8th nerve, with a condition known as "auditory neuropathy" [Berg et al., 2005; Rance et al., 1999; Starr et al., 1996]. Such babies will be missed when only TEOAE is used for screening. AABR on the other hand may miss babies with mild sensorineural or high frequency hearing loss. The choice of screening technology however, extends beyond auditory biophysics and neuro-physiology to matters of practicality, ergonomics and economics.

The world's first systematic UNHS programme conducted in the USA provided evidence in support of the choice of OAE as initial screen followed by AABR [Vohr et al., 1998].

A multi-centre study sponsored by the National Institutes of Health in USA to evaluate the accuracy of AABR, TEOAE and DPOAE confirmed all three tests as accurate and robust [Norton et al., 2000b]. Many studies in UK and USA have documented sensitivity and specificity in excess of 94% for TEOAE [Watkin, 2003; Mehl & Thomson, 2002]. The combination of TEOAE and AABR tests in a two-stage screening requiring AABR to be offered to those who failed an initial screen with TEOAE has been found to have the most favourable combination of specificity, sensitivity, acceptability, and high coverage in hospitals with a wide range of birth rates [Kennedy et al., 2005]. It is the preferred protocol for the National UNHS Programme in the UK and many state-mandated programmes in the USA [Johnson et al., 2005; Lin et al., 2005; Hall 3rd, Smith & Popelka, 2004; Vohr et al., 2001; Gravel et al., 2000; Kennedy et al., 2000]. In fact, some screening instruments have been designed with the capability to carry out two or three combination of TEOAE, DPOAE and AABR.

Notwithstanding its widespread application, a two-stage screening with TEOAE and AABR has been reported to miss about 23% of those with permanent mild hearing loss when a follow-up visit for repeat diagnostic evaluation occurs at about 9 months of age [Johnson et al., 2005]. This is partly attributable to the fact that current AABR technology is designed only to identify infants with moderate to profound hearing loss. It is therefore important to note this limitation in programmes with this protocol, especially when advising parents of babies with established risk factors who pass AABR.

2.2.5. Rationale for Universal Newborn Hearing Screening

Despite the availability of OAE and ABR, hearing screening of newborns was based on the JCIH ten risk factors in the early 1990s. However, it was observed that such targeted screening was not efficient and effective in the

early detection of babies with PCEHL as it was likely to miss about 50% of babies with moderate to profound hearing loss [Mauk et al., 1991; Watkin, Baldwin & McEnery, 1991]. Moreover, a significant proportion (38 – 60%) of infants with PCEHL did not exhibit any known risk factor(s) while some of the risk factors were not easily detectable at birth or shortly thereafter [Mozaria, Westerberg & Kozak, 2004; Sellars & Brighton, 1983]. This situation led to considerable delay in detection, usually beyond 12 months, for the vast majority of infants with PCEHL especially of mild-to-moderate degree [Mauk et al., 1991; Stein et al., 1990].

In the UK where a complementary Health Visitor Distraction Test (HVDT) was universally applied for screening infants aged 7 - 9 months, the age of identification of hearing loss ranged from 12 – 20 months [Davis et al., 1997; Watkin, Baldwin & Laoide, 1990]. A survey of nine European countries including UK by Martin et al [1981] in fact showed that hearing loss was suspected or identified in only 24% of children in the first year of life and in two-thirds by the age of 3 years. By implication, intervention was equally delayed beyond the first year of life with sub-optimal outcomes [Ramkalawan & Davis, 1992; Culberton & Gilbert, 1986; Davis et al., 1986; Blair, Peterson & Viehweg, 1985].

In 1988, Dr. Peter Watkin pioneered newborn hearing screening with OAEs at the Whipps Cross Hospital, in the UK [Watkin, Baldwin & McEnery, 1991]. This was believed to be the first routine UNHS with OAEs in the world and was followed by the first-ever controlled trial of universal newborn screening worldwide in the Wessex region from 1993-1996 [Wessex Trial Group, 1998]. Following the encouraging results from these programmes and the need for a thorough evaluation of various screening options as well as service requirements [Curnock, 1993], the UK National Screening Committee (NSC) commissioned a reappraisal of the HVDT as a universal infant screening at 7-9 months and the ad hoc targeted screening of high-risk babies in some health districts. In their systematic review, Davis et al [1997] concluded that UNHS had a lower running cost and much lower cost per child detected than

HVDT. It therefore recommended the introduction of UNHS, supplemented by selective HVDT at age 7 months to catch those missed by UNHS or those who developed PCEHL after birth. Further evidence was provided by the Wessex Universal Neonatal Hearing Screening Trial Group through a controlled trial of the effectiveness of UNHS over the HVDT [Wessex Trial Group, 1998]. These reports significantly stimulated the introduction of pilot programmes by the UK National Screening Committee in 1999, preparatory to the nationwide implementation of UNHS in 2001 [Bamford, Uus & Davis, 2005; Davis & Hind, 2003]. This development did not preclude on-going attempts by other researchers to find practical and inexpensive tools for identifying hearing loss in pre-school children in the UK [Bellman, Mahon & Triggs, 1996].

In 1998, the European Consensus Statement on Neonatal Hearing Screening was also released [Lutman & Grandori, 1999]. It noted that neonatal screening in maternity hospitals was more effective and less expensive than the conventional behavioural screening at 7-9 months; and that targeted neonatal screening in conjunction with the 7 – 9 month behavioural screening was more expensive and less effective than UNHS. It therefore recommended that neonatal hearing screening should be considered as the first part of a programme for habilitating hearing-impaired children. This Statement inspired the introduction of pilot hospital-based UNHS in many European countries.

In the USA, the National Institutes for Health Consensus Conference on Newborn Hearing Screening in 1993 recommended the screening of all infants for hearing impairment within the first three months of life [NIH, 1993]. This was followed by JCIH recommendations in 1994 for the detection of hearing loss by three months of age and intervention by six months [AAP, 1995]. However, some concerns were expressed about the feasibility, risk and cost-effectiveness of such a programme on the grounds that the available research evidence and experience for such large-scale implementation was not convincing [US Preventive Services Task Force, 1996; Bess & Paradise, 1994]. The emerging evidence from large-scale UNHS hospital-based

programmes between 1995 and 1998 substantially addressed most of these reservations and challenges [Finitzo, Albright & O'Neal, 1998; Mason & Herrmann, 1998; Mehl &Thomson, 1998; Vohr et al., 1998; Barsky-Firkser & Sun, 1997; Maxon et al., 1995]. For instance, Vohr et al [1998] pioneered a state-wide UNHS in Rhode Island from 1993 to 1996 in which 99% of the 53,121 live births were successfully screened before hospital discharge with a 10% referral rate thus demonstrating the feasibility of UNHS and the prospects of lower referral rates with improvements in screening technologies and experience. Screening was conducted by trained technicians, nurses and nurses' aides. The mean age of hearing loss confirmation was reduced significantly from 8.7 months in the first year of the programme (1993) to 3.5 months by the fourth year. Similarly, Mehl & Thomson [1998] reported the outcomes of UNHS in 26 hospitals in Colorado in which 41,796 newborns were successfully screened between from 1992 to 1996 with a cumulative false positive rate of about 6%. Over 80% of the screening was conducted by volunteers, technicians and nurses rather than audiologists. The cost of screening a baby ranged from US$18 to US$33 with a mean cost of US$25. Compared to other newborn screening programmes for say phenylketonuria and hypothyroidism, UNHS was found to be more cost-effective based on the cost per case detected. Intervention services including the fitting of hearing aids were initiated between 3 to 6 months for about 80% of infants confirmed with hearing loss. In addition, the landmark publication by Yoshinaga-Itano and colleagues [1998] demonstrated that children with all degrees of hearing loss who received intervention services prior to six months of age had speech, language and cognitive skills comparable to their normal hearing peers when tested at age 3 years.

Based on the evidence from these US studies and from the UK reports, the Task Force on Newborn and Infant Hearing of the American Academy of Pediatrics in 1999 endorsed the implementation of UNHS and outlined the parameters for evaluating its effectiveness. Physicians in each birthing hospital were required to direct such a programme [AAP, 1999]. Subsequent reports showed better developmental outcomes for hearing-impaired children

born in hospitals with UNHS programmes than those born in hospitals without [Yoshinaga-Itano, 2003; Yoshinaga-Itano, Coutler & Thomson, 2001]. In addition, substantial reduction in the average age of identification and intervention for PCEHL was observed in the US from the more than 2 years of age previously reported before UNHS to less than 3 months [Harrison, Roush & Wallace, 2003].

In 2002, the JCIH set out the following principles for any UNHS programme:

1. All infants regardless of their place of birth should be screened in the first month of life.

2. All infants failing the screening test(s) should undergo appropriate medical and audiological evaluations necessary for the confirmation of hearing loss (if any) before 3 months of age.

3. All infants with confirmed permanent hearing loss should receive family-oriented and culturally appropriate intervention services before 6 months of age.

4. All infants who pass the screening test(s) but have risk indicators for auditory disorders and/or speech and language delay should receive on-going surveillance with audiological assessment every 6 months until age 3 years.

The JCIH further recommended the following quality benchmarks for hospital-based infant hearing screening programmes:

1. A minimum screening coverage of 95% of eligible infants must be achieved within the first 6 months of initiating the programme.
2. A maximum referral rate of 4% should be achieved by the first year anniversary of the programme.
3. A minimum return-for-follow-up rate of 70% for diagnostic tests.
4. Confirmation of hearing loss by 3 months of age and commencement of intervention by 6 months of age.

In the UK, the national targets were initially set to detect 80% of bilateral congenital hearing loss within the first year of life, and 40% by 6 months of age [National Deaf Children Society (NDCS), UK, 1994]. Hearing aids were to be fitted within four weeks of confirmation of hearing loss where appropriate. At the commencement of the National Newborn Hearing Screening Programme (NHSP) in 2001 the following targets were set [Davis & Hind, 2003]:

1. Identify 90% of children with bilateral moderate to profound permanent childhood hearing loss within 8 weeks of age and 100% by 24 weeks of age.
2. Begin an agreed habilitative programme with the family and child as soon as possible following confirmation of permanent hearing loss.
3. Provide follow-up services for children with unilateral or mild hearing loss and those at risk of progressive hearing loss at an appropriate age.

These targets were subsequently revised to the current standards as follows:

1. To achieve a screen coverage of 95-100% by 6 months of age.
2. First (OAE) screen should take place within 10-24 days.
3. Second (OAE) screen (if required) should take place within 7 days from the first screen.
4. AABR screen should take place within 5 weeks of birth.
5. For hospital-based programmes, 1st and 2nd screen should be done before hospital discharge.
6. Missed babies should be screened as outpatients or in the community by 4 weeks.

Recognising the limitations of hospital-based programmes that screened babies within 48 hours of birth in detecting certain categories of PCEHL and based on the emerging evidence for large-scale epidemiological studies in USA and the UK [Cone-Wesson et al., 2000; Norton et al., 2000a; Fortnum & Davis 1997], the JCIH revised its 1994 risk factors for infants less than 29 days and for infants 29 days to 24 months old who may develop hearing loss as a result of adverse post-natal conditions or as a consequence of essential medical interventions in the treatment of illness (Table 2-3).

The risk factors were intended primarily for monitoring infants with PCEHL who were missed by the hospital-based hearing screening or those who were likely to have progressive or delayed-onset PCEHL despite passing neonatal hearing screening [Barbi et al., 2003; Cunningham et al., 2003; Williamson et al., 1992; Fowler et al., 1999]. In addition, it was suggested that the risk factors could be used in settings where UNHS was not immediately practicable, particularly in developing countries [JCIH, 2000; Mencher & Mencher, 1999; Gell et al., 1992].

Some researchers however, still argued that targeted screening was more cost-effective than UNHS even for developed countries [Kileny & Lesperance, 2001] and that some of its limitations could be possibly addressed by the identification of additional risk factors not currently listed by JCIH [Kountakis et al., 2002]. Beyond this debate, there is little doubt that the knowledge of these risk factors is of clinical relevance to health professionals particularly in determining the natural history of these conditions and the risk of reoccurrence [Vohr et al., 2000].

Table 2-3. JCIH Risk Factors for Hearing Loss

JCIH	Risk Factors for Hearing Loss
1990 & 1994 *Birth to 28 Days*	• *Family history of sensorineural hearing loss.* • *In-utero infections such as rubella, cytomegalovirus, syphilis, toxoplasmosis and herpes.* • *Cranio-facial anomalies.* • *Birth weight less than 1,500g (3.3lbs).* • *Hyperbilirubinaemia at levels requiring exchange transfusion* • *Ototoxic medications.* • *.Bacterial meningitis.* • *Birth asphyxia with Apgar score 0-4 at 1 minute or 0-6 at 5 minutes* • *Mechanical ventilation lasting five days or more.* • *Stigmata or other findings associated with a syndrome known to include a sensorineural and/or conductive hearing loss.*
2000 *Birth to 28 Days*	• *Family history of sensorineural hearing loss.* • NICU admission greater than 48 hours • *Stigmata or other findings associated with a syndrome known to include a sensorineural and/or conductive hearing loss.* • *.Cranio-facial anomalies.* • *In-utero infections such as rubella, cytomegalovirus, syphilis, toxoplasmosis and herpes.*
2000 *29 days to 2 years*	• *Parental/caregiver's concern regarding hearing, speech, language and or developmental delay* • *Family history of sensorineural hearing loss.* • *Stigmata or other findings associated with a syndrome known to include a sensorineural and/or conductive hearing loss or Eustachian tube dysfunction.* • *Postnatal infections associated with sensorineural hearing loss including bacterial meningitis* • *In-utero infections such as cytomegalovirus, herpes, rubella, syphilis and toxoplasmosis* • *Neonatal indicators especially hyperbilirubinaemia at levels requiring exchange blood transfusion* • Syndromes associated with progressive hearing loss, such as neurofibromatosis, osteopetrosis, and Usher's syndrome • *Neurodegenerative disorders, such as Hunter syndrome, or sensory motor neuropathies, such as Fredreich's ataxia and Charcot-Marie-Tooth syndrome.* • *Head trauma* • *Recurrent or persistent Otitis Media for at least 3 months*

2.3. Early Hearing Detection in Developing Countries

2.3.1. Types of Early Hearing Detection Programmes

The four generic options for the detection of PCEHL are depicted in Figure 2.14.

	Hospital-based	Community-based
Targeted	**TH**	**TC**
Universal	**UH**	**UC**

Figure 2.14. Infant Hearing Screening Models

They consist of Targeted or Universal Hearing Screening of newborns in hospitals before discharge and Targeted or Universal Hearing Screening of infants at community health centres during visits for child health services such as routine immunisations [Lin et al., 2004; Owen, Webb & Evans, 2001; Bantock & Croxson, 1998; McPherson et al., 1998]. The main characteristics of these options are examined as follows:

2.3.1.1. Hospital-based Screening Models

Hospital-based programmes provide the largest captive population for newborn hearing screening in countries or communities where a significant number of babies are born in hospitals. Hospitals with birthing facilities where infant hearing screening can be conducted may range from publicly owned tertiary or teaching hospitals [Mukari, Tan & Abdullah, 2006; Khairi et al., 2005; Ng et al., 2004], specialist maternity hospitals [Flynn et al., 2004; Bailey et al., 2002], general or district hospitals [Quintos et al., 2003; Watkin, Baldwin & McEnery, 1991] or community hospitals [Attias et al., 2006] to private hospitals [Swanepoel et al., 2007; Yee-Arellano, Leal-Garza & Pauli-

Muller, 2006; Chapchap & Segre, 2001]. Most hospital-based programmes aim to screen babies usually by 48 hours after birth and preferably before hospital discharge. Screening before the first 24 hours of birth is not encouraged because of high false positive rates associated with vernix plugs or amniotic fluid in the babies' ears except if it is unavoidable due to early discharge [Doyle et al., 2000; Maxon et al., 1997; McNellis & Klein, 1997]. The screening programme could be targeted for babies with pre-determined risk factors (TH) or offered to all babies (UH). Although most hospital-based programmes are stand-alone, it may be possible to link them to existing hospital-based programmes like the Baby-Friendly Hospital Initiative (BFHI) [Olusanya, Luxon & Wirz, 2004]. The BFHI is a global WHO/UNICEF-sponsored effort to encourage exclusive breast-feeding from birth to the age of 6 months. It has a unique advantage of providing regular contact between health-care professionals and nursing mothers to promote breast-feeding through a series of ten steps. Breastfeeding is culturally acceptable in many developing countries and has made the BFHI campaign popular among women [UNICEF, 2006]. Many hospitals (including privately-owned ones) and community health centres have incorporated this programme into their ante-natal clinics. UNHS may have a high prospect for a good coverage in the target population if included in this campaign.

2.3.1.2. Community-based Models

In many developing countries a significant number of births occur outside regular hospital facilities. Table 2-4 shows that the proportion of deliveries with skilled birth attendants (such as doctors, nurses and trained midwives), which is an index for hospital deliveries, varies from as low as 13% in Bangladesh to as high as 97% in Malaysia [UNICEF, 2006].

Deliveries at home and in traditional maternity centres often account for the significant number of deliveries outside hospitals [Blum, Sharmin & Ronsmans, 2006; Borghi et al., 2005; Osrin et al., 2002]. Unfavourable reports on the activities of unskilled birth attendants in communities where a large proportion of births occur outside regular health facilities has put into

question the safety and effectiveness of deliveries at traditional maternity centres or private homes [Blum, Sharmin & Ronsmans, 2006; Sreeramareddy et al., 2006; WHO, 2004b].

Table 2-4. Live Births with Skilled Birth Attendants in Selected Countries

Country [Region]	Annual Live Births ('000) [2004]	% with SBA* [1996-2004]
Bangladesh [South Asia]	3.738	13
Pakistan [South Asia]	4,729	23
Nigeria [Sub-Sahara Africa]	5,323	35
India [South Asia]	26,000	43
South Africa [Sub-Sahara Africa]	1,093	84
Saudi Arabia [Middle-East]	665	91
Brazil [Latin America]	3,728	96
Malaysia [East Asia]	549	97

*Skilled Birth Attendants. Source: UNICEF, 2006

However, home deliveries are not always or necessarily unsafe especially for healthy and low risk women [Sreeramareddy et al., 2006; Ackermann-Liebrich et al., 1996; Wiegers et al., 1996]. They are inevitable for some women who have no access to regular health facilities and may be preferred by a significant number of women for a variety of reasons even in urban areas well served with hospital facilities [Ekele & Tunau, 2007; Koblinsky et al., 2006; Sreeramareddy et al., 2006; Fernandez, Mondkar & Mathai, 2003].

Asking parents to go or return to the hospital after delivery/discharge specifically for an "unfamiliar UNHS" may result in a high rate of defaulters as recently demonstrated by Kolski et al [2007]. Similarly, it may be difficult to convince mothers to leave home or visit a location other than where they

delivered solely for the purpose of infant hearing screening. Even though home-based infant hearing screening has been experimented in some developed countries [Owen, Webb & Evans, 2001; Oudesluys-Murphy & Harlaar, 1997], it is perhaps impracticable to contemplate such an option in a developing country because of the enormous logistical challenges it may entail [Blum, Sharmin & Ronsmans, 2006]. It is probably more practicable and expedient therefore to target existing healthcare facilities which have been reasonably integrated into primary healthcare services and are capable of attracting infants regardless of their places of birth. This approach has been used to adapt the existing well-established programmes to incorporate new child health initiatives in some countries like Tanzania and Ethiopia [Schellenberg et al., 2001; Edmunds et al., 2000]. Current platforms that could be valuable for the introduction of UNHS based on this approach are examined briefly:

2.3.1.2.1. Expanded Programme on Immunisation

Expanded Programme on Immunisation (EPI) is a global initiative of UNICEF to deliver vaccinations against tuberculosis, diphtheria, pertussis, tetanus, measles and more recently, hepatitis B to infants. A typical immunisation schedule in a developing country is shown in Table 2-5 [WHO, 2002b].

Table 2-5. Typical National Immunisation Schedule in Developing Countries

Vaccine	Recommended age for vaccination				
	Birth	6 weeks	10 weeks	14 weeks	9 months
Bacille Calmette-Guérin (BCG)	x				
Oral polio*	x	x	x	x	
Diphtheria-pertussis-tetanus (DPT)		x	x	x	
Hepatitis B*		x	x	x	
Haemophilus influenza type b*		x	x	x	
Yellow fever*					x
Measles					x

* Not provided in, or applicable to all countries. Source: WHO, 2002b

The vaccines are given at birth (Bacille Calmette-Guérin - BCG), before the age of four months (dipththeria-pertussis-tetanus-DPT1, 2, 3; Oral Polio1, 2, 3; Hepatitis B1), and at 9 months (measles; yellow fever). BCG immunisation often records the highest uptake of all the vaccinations administered in the first year in the developing world based on data from UNICEF and WHO (Figure 2.15).

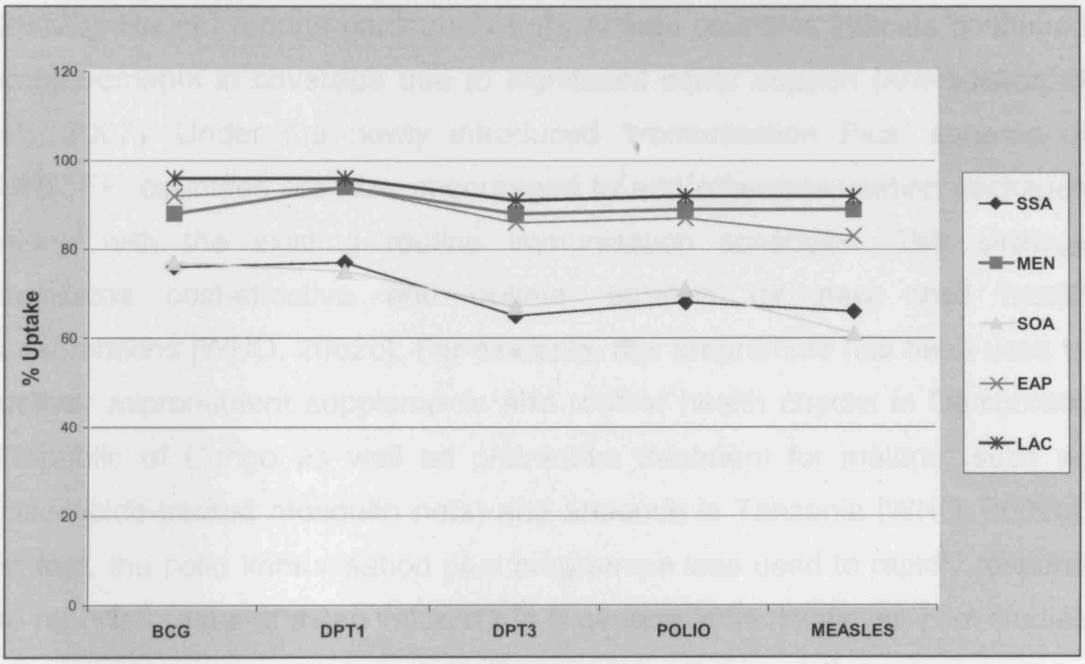


Figure 2.15. Expanded Programme on Immunisation in Developing Countries

Key: Sub-Saharan Africa (SSA); Middle East and North Africa (MEN); South Asia (SOA); East Asia and Pacific (EAP); Latin America and the Caribbean (LAC).

For instance, BCG was given at birth or shortly thereafter in 157 countries in 2002 with a coverage above 90% in 101 countries and below 60% in only nine countries [Trunz, Fine & Dye, 2006]. It has also been documented as a highly-cost effective intervention which reinforces its potential advantage for an infant hearing screening programme [Trunz, Fine & Dye, 2006]. However, the age at which babies are presented for the various immunisations varies widely within and across communities [Swanepoel, Hugo & Louw, 2006]. Nonetheless, the repeated visits for the multi-dose vaccines often spaced

four weeks apart and completed on or before the age of six months offer a good opportunity for the promotion of UNHS and subsequent follow-up of positive cases [Bantock & Croxson, 1998; McPherson et al., 1998].

The experiences in many countries suggest strongly that immunisation programmes are not only the most established child health intervention globally, but are also the best supported, through multilateral collaborations, well beyond the capacities of individual countries [UNICEF, 2006; WHO, 2002b]. Recent reports particularly from African countries indicate continued improvements in coverage due to significant donor support [Arevshatian et al., 2007]. Under the newly introduced "Immunisation Plus" scheme of UNICEF, countries are now encouraged to add other intervention packages along with the existing routine immunisation schedules. This strategy facilitates cost-effective and optimal uptakes for new child health interventions [WHO, 2002b]. For example, this programme has been used to deliver micronutrient supplements and routine health checks in Democratic Republic of Congo as well as preventive treatment for malaria (such as insecticide-treated mosquito nets) and anaemia in Tanzania [WHO, 2002b]. In fact, the polio immunisation plus programme was used to rapidly respond to reported cases of avian influenza in Nigeria in 2006. However, pilot studies are necessary to establish the effectiveness of this platform for any new intervention as prevailing conditions in each country may produce less than optimal outcomes for some services [Chandramohan et al., 2007]. Moreover, marked disparities may exist is some countries between rural and urban areas which must be considered [Agarwal, Bhanot & Goindi, 2005; Atkinson & Cheyne, 1994].

2.3.1.2.2. Integrated Management of Childhood Illness

The Integrated Management of Childhood Illness (IMCI) is a WHO/UNICEF strategic initiative originally designed as an integrated case management approach to the five most important causes of childhood mortality - acute respiratory infections, diarrhoea, measles, malaria and malnutrition [Shah & Sachdev, 1999]. The key objectives of the strategy are

to reduce death, the frequency and severity of illness and disability. Apart from the five fatal diseases, the generic guidelines and Adaptation Guide have identified ear disorders as one of the conditions to be addressed. By its design, the IMCI strategy makes allowance for the content to be adapted to individual country's needs in terms of prevention of diseases, curative care interventions, and measures that promote healthy growth and development in children. IMCI has been implemented in many developing countries including Nigeria and it should also be possible to successfully implement a community-based programme for early hearing detection and intervention within this programme. Similarly, in India, a variant of IMCI - the Integrated Child Development Services (ICDS) - is reputed as the worlds largest with over 40,000 centres nation-wide reaching more than 34 million children aged 0-6 years and 7 million pregnant and nursing mothers [Kapil, 2002; UNICEF, India]. The ICDS has also been utilized to provide health and nutrition education, health services, supplementary food, and pre-school education.

2.3.2. Global Impetus for Early Hearing Detection in Developing Countries

Healthcare priorities in developing countries are often based on the policies and programmes of the United Nations and its agencies. Some of the current global initiatives of these institutions provide significant context and incentive for the introduction of early hearing detection programmes in developing countries. For example, two resolutions of the World Health Assembly (WHA48.9 & WHA58.23) urged member nations to take appropriate steps towards the early identification and management of children with hearing impairment and other disabilities while pledging the technical support of WHO to these countries [WHO, 2005b & 1995]. WHO's strategic document on the continuum of care for maternal and child health also makes explicit provision for the detection and management of hearing impairment from the first year of life until school age [WHO, 2003].

Similarly, UNICEF, UNESCO and the World Bank have early childhood programmes aimed at supporting children's survival, growth, development and learning from birth

to entry into primary school, especially for the most vulnerable and disadvantaged children. These programmes together acknowledge that it is more cost-effective to institute preventive measures and support for children with special needs early than to compensate for the disablement as they grow older [Young, 2007 & 2002; UNICEF, 2006 & 2002; UNESCO, 2006]. In addition, UN millennium development project constitutes the global agenda for the mobilisation and prioritisation of resources to the developing world till year 2015 [United Nations, 2005]. The first two of the millennium development goals (MDGs) are focused on the eradication of extreme poverty and the completion of primary education by children everywhere. It has been demonstrated that failure to detect and manage PCEHL through UNHS in developing countries will significantly undermine the realisation of these two goals [Olusanya, Ruben & Parving, 2006]. These initiatives have also stimulated a growing advocacy for the globalisation of infant hearing screening programmes [Olusanya et al., 2007].

2.3.3. Progress towards EHDI in Developing Countries

Typical examples of ongoing pilot programmes on infant hearing screening in the developing world are summarised in Table 2-6 [Olusanya et al., 2007]. The earliest reported project using electrophysiological tests in a developing country was perhaps initiated in India in 1986 among high risk infants [M. Jayaram, personal communication, September 14, 2005]. Although Ansari (2004) reported an unpublished project on newborn hearing screening in1984 there was no description of the screening methods. The second oldest screening programme is in Brazil and dates back to 1988 [Chapchap & Segre, 2001]. There are over 237 screening sites across many states in this country and the programme is by far the largest in any developing country.

As observed with pilot programmes in other developing countries, healthcare services are concentrated in urban areas presumably because of accessibility to the limited available services. In Oman, universal newborn hearing screening is currently offered routinely nationwide after prior pilot studies in various regions of the country [Khandekar et al., 2006]. This is perhaps the first developing country with a national programme on newborn hearing screening.

Table 2-6. Pilot Infant Hearing Screening Programmes in Developing Countries

Country [Reference]	Year Started; (Location)	Setting/ Coverage	Protocol	Total Screened [Duration]
South Africa [Swanepoel et al., 2006]	2003 (Pretoria)	Hospital, MCC: UNHS	OAE	510* [5 months]
India [http://healthlibrary.com]	2003 (Kochi)	Hospitals: TNHS	OAE	2,500* [N/A]
Pakistan [Ali et al., 2000]	1999 (Lahore)	Hospitals: UNHS	OAE	756* [N/A]
Saudi Arabia [Habib & Abdelgaffar, 2005]	1996 (Jeddah)	Hospitals: UNHS	OAE	11,986* [96months]
Iran [Masoud et al., 2006]	2002 (Tehran, Mashad)	Hospitals: UNHS	OAE	16,000**+ [6 months]
Qatar [Bener et al., 2005]	2003 (Doha)	Hospitals: UNHS	OAE	2,800* [11 months]
Jordan [Attias et al., 2006]	2001 (Multiple Cities)	Hospitals, MCC: UNHS	OAE	8,251** [N/A]
Oman [Khandekar et al., 2006]	2003 (Multiple cities)	Hospitals, MCC: UNHS	OAE, AABR	21,387** [12 months]
China [Xu, Zhang & Du, 2006]	2000 (Nanjing)	Hospitals, UNHS	OAE,AABR	8,800* [60 months]
Hong Kong [Ng et al., 2004]	1998 (Hong Kong	Hospital , UNHS	OAE, AABR	1,064* [5 months]
Taiwan [Lin et al., 2002]	1998 (Taipei)	Hospitals: UNHS	OAE, AABR	6,765* [24 months]
Malaysia [Mukari et al., 2006]	2000 (Kuala Lumpur)	Hospitals: UNHS	OAE	4,437* [11 months]
Philippines [Quintos et al., 2003]	2000 (Manila)	Hospitals: UNHS	OAE	406* [12 months]
Singapore [Low, Pang & Ho, 2005]	2002 (Singapore)	Hospitals: UNHS	AABR, OAE	36,095** [24 months]
Brazil [Chapchap & Segre, 2001]	1996 (Sao Paulo)	Hospitals: UNHS	OAE	4,196* [36 months]
Mexico [Yee-Arellano et al., 2006]	2005 (Mexico City)	Hospitals: TNHS, UNHS	OAE , AABR	3,066* [24 months]

MCC= Maternal and child health clinic. N/A= Not available. (*Single screening site, **Multi-screening sites)
TNHS= Targeted Newborn Hearing Screening; UNHS= Universal newborn hearing screening;
OAE= Otoacoustic emissions; AABR=Automated auditory brainstem response;
NHS2006= NHS Conference, Lake Como, Italy, May 31 – June 1, 2006 [Abstracts]

A national initiative towards screening almost half of the babies born in Chile based on risk factors has just been implemented by the Chilean Ministry of Health (http://www.press.hear-it.org). Pilot studies have also been conducted in 28 of the 30 provinces in Iran [Masoud et al., 2006] and the number of babies screened varied from 406 in Philippines [Quintos et al., 2003] to 36,095 in four hospitals in Singapore [Low, Pang & Ho, 2005]. The duration of the reported data ranged from 5 months in South Africa [Swanepoel, Hugo & Louw, 2006] and Hong Kong [Ng et al., 2004] to 60 months in China [Xu, Zhang & Du, 2006]. It is pertinent to mention that these pilot programmes are still on-going in most of these countries.

Overall, screening protocols consist of both universal and targeted approaches in which otoacoustic emissions (OAE) and/or automated auditory brainstem response (AABR) techniques are employed. The targeted screening is commonly based on the risk factors recommended by the JCIH [2000].

The majority of screening programmes are hospital-based except in South Africa, Taiwan and Hong Kong (and to a limited extent in China, Jordan and Oman) where community-based programmes have been reported [Attias et al., 2006; Khandekar et al., 2006; Lam, 2006; Swanepoel, Hugo & Louw, 2006; Ng et al., 2004; Lin et al., 2002]. Community-based programmes are typically implemented during routine immunisation clinics at community health centres. Preliminary results from South Africa confirm that infant hearing screening at immunisation clinics is feasible and worthwhile in developing countries [Swanepoel, Hugo & Louw, 2006], although more reports are needed from other regions.

Screening protocols and technologies differ from country to country and may account for variations in screening performance. For instance, a screening protocol consisting of a first–stage DPOAE and a high frequency probe tone (1 kHz) tympanometry for infants from birth to 12 months of age was used in the screening programme in South Africa. A planned second-stage screen

with AABR was discontinued because of practical difficulties with administering the test to babies when they are older than three months. Rather, the first-stage protocol was repeated for subsequent stage(s). Only 40% of those scheduled for the second-stage screen returned, which could be due to the timing of follow-up visits outside a routine immunisation schedule.

Although the default rates from South Africa and some other developing countries are initially high, they are comparable to reported rates in the early stages of most UNHS programmes in the developed world [Korres et al., 2006; Mehl & Thomson, 2002; Vohr et al., 1998]. Notwithstanding the challenges highlighted in the hospital and community settings, these reports together would suggest that infant hearing screening is feasible in both settings in developing countries [Olusanya et al., 2007].

2.4. Framework for Infant Hearing Screening in Nigeria

Screening as a public healthcare service must satisfy a number of criteria first set out by Wilson & Jungner [1968] and more recently applied to non-communicable diseases by Strong and colleagues [2005]. Since screening procedures are not always beneficial, an essential scientific and ethical requirement that must be satisfied before any screening programme can be introduced is that the benefits of screening should outweigh the potential risks or harm associated with the screening procedure [Raffle, 2006; Strong, et al., 2005; National Screening Committee (UK), 2003].

Within the framework for screening for non-communicable diseases and the ethical demand of the "rule of rescue" for a health condition that is associated with an optimal intervention that is time-bound, the following four core principles or criteria are necessary before an infant hearing screening programme can be justified for Nigeria [Olusanya, Luxon & Wirz, 2006b & 2005b; National Screening Committee (UK), 2003; Landman & Henley, 1999; Hadorn, 1991]:

- The condition must be a significant health problem;
- The screening test must be simple, safe, reliable and valid;
- The treatment or intervention must be effective and available;
- The benefits from the screening programme must outweigh the risk of harm.

It is also useful to establish the economic implications of such a programme for an individual and the society to ensure that it falls within an acceptable cost-effective range before being widely implemented as a public healthcare service [American Academy of Pediatrics, 1999].

2.4.1. The Significance of PCEHL in Nigeria

Although several studies among children in deaf schools or in hospital-based population have been conducted, the epidemiology of PCEHL in Nigeria is still unknown [Dumade et al, 2007; Lasisi , Ayodele & Ijaduola, 2006; McPherson & Swart, 1997; Chukwuezi, 1991; Obiako, 1987; Holborow, Martison & Anger, 1982; Ijaduola, 1982]. Nonetheless, these studies provide some insights on PCEHL in Nigeria in which aetiological factors such as infections, bilirubin encephalopathy, ototoxicity and hypoxic toxaemia have been implicated. For instance, conditions such as congenital syphilis, rubella, hypoxia, cerebral palsy, neonatal jaundice, kernicterus, maternal rubella, sickle cell disease, and familial deafness have been reported. However, a common limitation of these studies is the frequent reliance on medical history as the basis for aetiological classification which results in a significant group of about 30-35% with unknown aetiologies.

Genetic factors usually account for about one-third of the incidence of PCEHL, environmental infections or toxins (including intra-uterine infections such CMV and rubella) for another one-third while the final one-third is classified as unknown. Improved identification of aetiological factors due to recent advances in the genetic epidemiology of deafness is likely to reduce the size of the "unknown category" over time [Morton & Nance, 2006; Smith, Bale & White, 2005; White, 2004]. Available data among school children indicate

prevalence rate of 33 to 89 per 1000 for permanent hearing loss in this age group [National Ear Care Programme, Nigeria (NECP), 2002; Ogisi & Amu, 1990]. A recent hospital-based study in Lagos also found permanent hearing loss as the commonest communication disorder among children presenting at weekly ENT clinics [Somefun et al., 2006].

In the UK, for example, about 840 children are born every year with significant hearing impairment and it is estimated that one in every 1,000 children become deaf by the age of 3 years [Royal National Institute for the Deaf (RNID), UK, 2003]. Recent estimates from on-going UNHS programmes in developed countries suggest that the prevalence of permanent congenital hearing impairment ranges from 2 to 4 per thousand live births [Smith, Bale & White, 2005; White, 2004]. This estimate takes into account bilateral or unilateral permanent hearing loss greater than 30 dB HL. Using the projected prevalence of 6 per 1000 for developing countries [Olusanya, Ruben & Parving, 2006], an estimated 31,800 babies are likely to be born annually with significant permanent hearing loss in Nigeria and about 30,623 will live beyond their fifth birthday for an average of 43 years [UNICEF, 2006]. This projection for the incidence of PCEHL may in fact be an underestimation when one considers the prevailing risk factors for disabling and permanent childhood hearing impairment associated with the poor health and socio-economic conditions in Nigeria as with many developing countries [Dunmade et al., 2007; Dawodu, 1998; Chukuezi, 1991; Ijaduola, 1982]. For example, birth asphyxia from prolonged obstructed labour has been reported in local clinical practice [Njokanma, Sule-Odu & Akesode, 1994; Anate, 1993; Akpala, 1993]. Birth asphyxia is often described clinically by poor Apgar scores despite their limitation in predicting neurological outcome, a use for which they were originally not meant to serve [Casey, McIntire & Leveno, 2001; Wolf et al., 1998; Misra et al., 1994; Nelson & Ellenberg, 1981]. Established obstetric practices in Nigeria include routine recording of Apgar scores at one and five minutes. Apgar score of ≤ 4 in 1 minute or ≤ 6 in 5 minutes is one of the JCIH lists of significant risk factors for PCEHL.

Existing literature from various parts of the country suggests that neonatal hyperbilirubinaemia either as bilirubin encephalopathy or kernicterus still constitutes a significant disorder among Nigerian infants [Ahmed, Yakubu & Hendrickse, 1995; Owa & Dawodu, 1990; Sodeinde et al., 1995; Olowe & Ransome-Kuti, 1981]. More recently, neonatal hyperbilirubinaemia which may cause significant anomalies of the mitochondrial function has been linked with auditory neuropathy in the developed countries [Shapiro, 2003]. Auditory neuropathy is a recently described specific condition characterised by intact sensory outer hair cell function but auditory nerve dysfunction. This complex, multi-factorial clinical entity with variable outcomes can be difficult to detect when hearing screening is limited to TEOAE only or when TEOAE is the first stage screen.

Other documented risk factors include febrile illness [Ibekwe, 1998], sickle cell anaemia [Mgbor & Emodi, 2004; Omotade et al., 1998; Odetoyinbo & Adekile, 1987; Ogisi & Okafor, 1987], intra-uterine infections [Oyemade & Odelola, 1985], and ototoxicity from chloroquine and aminoglycosides [Obiako, 1985; Mukherjee & Mukherjee, 1979]. In addition, but relatively rare, congenital malformations together with or without syndromes have been described in local studies [Adeyemo, Gbadegesin & Omotade, 1997; Adeyokunnu, 1982; Mcmoli & Ijaduola, 1981; Amoni & Abdurrahman,1979].

Other adverse perinatal conditions that may be associated with PCEHL include, neonatal seizures [Obi, Ejeheri & Alakija, 1994; Okoji, Peterside & Oruamabo, 1993; Airede, 1991], small-for-gestational-age [Okoji & Oruamabo, 1992], neonatal sepsis [Akindele & Rotilu, 1997], prolonged rupture of membrane [Owa et al., 1990], congenital malaria [Runsewe-Abiodun, Ogunfowora & Fetuga, 2006; Obiajunwa, Owa & Adeodu, 2005; Egwunyenga et al., 1996; Akindele, Sowunmi & Aboweyere, 1993] and young maternal age [Ampofo, Otu & Uchebo, 1990].

Studies elsewhere have shown that one or more of these risk factors are present even when they are not manifested in over two-thirds of children

detected with PCEHL [Kountakis et al., 2002; Vohr et al., 2000]. Against this backdrop, PCEHL is unlikely to be a less significant health condition in Nigeria compared to countries with mandatory screening programmes for infant hearing loss.

2.4.2. Quality of Available Screening Options

There is currently no form of routine or systematic screening for hearing loss at any level of healthcare delivery in Nigeria. As in the developed world, parents are often the first to suspect the existence of a hearing loss as a result of a child's inattention, erratic response to sound or speech delays [Prendergast, Lartz & Fiedler, 2002; Watkin, Baldwin & Laoide, 1990; Parving, 1984]. This was evident in a recent study conducted in the two largest public schools for the deaf in Lagos with a total enrolment of 429 pupils (mean age: 10.3 years) [Olusanya, Luxon & Wirz, 2005a]. Parents were predominantly the first to suspect or detect hearing difficulty in their children (81%), and this occurred mostly in the second year of life. Only 12% of parents suspected hearing difficulty within the first six months of life. The commonest mode of detection was the child's failure to respond to sound (49%). Speech and language defects or unintelligible speech were least associated with hearing difficulty by parents (1%).

Parental concern may be slow or non-existent for children with mild, unilateral or fluctuating hearing loss, except in those in whom unusual psychosocial behaviours are observed. For instance, children with mild hearing loss would respond to loud sounds but have difficulty hearing soft sounds and speech, particularly when there is background noise. This partial ability to hear sounds may mislead parents in their judgement of whether or not their children are hearing impaired. A study showed that only a quarter of children with permanent hearing impairment were actually detected through parental concern even in the U.K. [Watkin, Baldwin & Laoide, 1990]. One study in Cyprus showed that only 40% of children with bilateral congenital impairment of 50 dB HL or greater were detected before the age of 24 months, while the mean age of detection overall was 44 months [Hadjikakou & Bamford, 2000].

Another study reported a mean age of suspicion of hearing impairment of about 38 months from a range of 18 months for profoundly hearing impaired children to 56 months for those with moderate impairment [Uus & Davis, 2000]. Understandably, the average age of identification was higher for milder degrees of hearing loss.

Further delays in diagnosis and intervention even with severe-to-profoundly deaf children have been linked to the failure by physicians to investigate hearing loss suspected by parents [Prendergast, Lartz & Fiedler, 2002; Parving, 1984]. Previously, it was not uncommon to find healthcare providers counsel that infants were too young to be tested or that they would outgrow the impairment thus unduly prolonging diagnosis and intervention. Often, hearing impairment was perceived as a developmental delay rather than a life-long developmental impairment. Unfortunately "delay" gives the impression that normal hearing will eventually be achieved as the disorder gradually disappears. But this is not the case for PCEHL, which has prompted some clinicians to caution against the indiscriminate use of the term "developmental delay" synonymously with "developmental impairment" [Bosley, 2005; Williams & Essex, 2004]. In one report (which may be regarded as outdated for the developed world but typical for a developing country), misdiagnosis by health personnel accounted for delayed identification in almost 30% of cases, following initial diagnosis such as mental retardation, aphasia, minimal brain damage, and learning disability [Hudgson, 1969]. These experiences are still common in Nigeria and some other developing countries.

Emerging evidence from studies on the cognitive development in high risk infants has shown that mother-infant-interaction and maternal responsiveness are crucial for positive outcomes [Smith, Landry & Swank, 2006; Wendland-Carro, Piccinini & Miller, 1999; Bornstein & Tamis-LeMonda, 1989; Olson, Bates & Bayles, 1984]. Even where intervention services are limited, parents, especially mothers, have intuitive skills to support children with special needs [Meadow-Orlans et al., 2004]. Hence mothers want and need to know early if their child has special needs [Hergils & Hergils, 2000;

Stuart, Moretz & Yang, 2000; Luterman & Kurtzer-White, 1999; Watkin, Beckman & Baldwin, 1995].

Since OAE and AABR are more reliable for the accurate and timely detection of PCEHL and are currently successfully used in other developing countries like South Africa and Brazil, there is evidence to suggest that these tests would be equally suitable and acceptable to parents in Nigeria particularly among those in an urban community. For instance, in a recent survey among mothers and mothers-to-be in Lagos, parental attitude towards newborn hearing screening was highly favourable [Olusanya, Luxon & Wirz, 2006a] and this was corroborated by the views of parents of deaf children in the same city [Olusanya, Luxon & Wirz, 2005a]. In addition, OAE and AABR are likely to be acceptable because they are non-invasive, quick to administer and do not require the use of drugs or injections.

2.4.3. Effectiveness of Early Intervention Options

The effects of PCEHL and the developmental needs of a child with PCEHL are varied and distinct. The scope and goals of any intervention must be matched with the specific needs of the child (and of course, the parents). The effectiveness of intervention options must be measured in relation to the specific goal of the intervention programme. PCEHL adversely affects four crucial developmental domains in early childhood as depicted in Figure 2.1 along with literacy skills, education and vocational attainment. Optimal speech and language development have very strong linkages with most of the remaining skills and for that reason it has often been the primary focus of many early intervention programmes. Nonetheless, intervention can be targeted independently at achieving results in the other areas [Carney & Moeller, 1998]. For instance, early intervention targeting sensory and perceptual skill development may include but not restricted to the provision of amplification devices. Intervention goal for language development may focus on enhancing parent/infant communication in the chosen or most feasible communication modality and developing verbal and reasoning skills to support literacy attainment [Schick, Marschark & Spencer, 2006; Spencer &

Marschark, 2006]. Similarly, intervention may simply focus on increasing reading and literacy skills as well as optimising overall educational achievement with a specific language base [Ramey & Ramey, 2006]; or intervention in the psychosocial domain may seek to establish appropriate family understanding and acceptance of hearing loss, reduce family stress as the child develops and improve social and emotional development throughout the school years [Carney & Moeller, 1998].

In Nigeria, enrolment in the schools for the deaf presently constitutes the conventional option for children with PCEHL principally due to the considerable delays in detection and poor awareness among health professionals and parents. In one study, it was reported that doctors were most commonly consulted for help (77%), but that the majority of children (80%) were rarely provided with hearing aids because children were often considered too young to be fitted with these devices [Olusanya, Luxon & Wirz, 2005a]. The primary mode of intervention often suggested to parents, was enrolment into a school for children that are deaf where sign language is the sole mode of communication [Olusanya, Luxon & Wirz, 2005a]. Even then, only about 6% of the children were enrolled in the school by 6 years of age. Consequently, the reading and comprehension abilities of these children are about 50-60% of their chronological age by the time they leave school typically at the age of 18 years. From a recent study [Olusanya & Okolo, 2006] it is important to note that parents from lower socio-economic class in Nigeria who constitute the vast majority, were less likely to enrol their children in the schools for children that are deaf. This would suggest that majority of children with hearing loss from lower socio-economic classes were unlikely to receive any form of intervention for communication and would be more susceptible to abuse and neglect [Hibbard et al., 2007; Knutson, Johnson & Sullivan, 2004; Kvam, 2004; Togonu-Bickersteth & Odebiyi, 1985].

In addition, the use of traditional and unorthodox therapies such as medicinal plants and animal fat for deafness with doubtful efficacy is a common recourse

for parents in Nigeria as in other developing countries because of ignorance and superstitious beliefs [Andrade & Ross, 2005; Byford & Veenstra, 2004; Kiyaga & Moores, 2003; Lasisi & Ajuwon, 2002; Stephens, Stephens & Eisenhart-Rothe, 2000; Keith, 1988; Odebiyi & Togonu-Bickersteth, 1987].

However, these trends can be redressed with greater public awareness particularly among health professionals and parents on the range of current possibilities for the effective management of PCEHL. The choice of appropriate communication and educational modalities for children who are deaf and hard of hearing is still a subject of debate worldwide [Lynas, 2005; Gravel & O'Gara, 2003]. However, since majority of parents of children with hearing impairment use spoken language and would prefer to develop verbal communication with their children, efforts could be made to help parents achieve this goal as far as practicable failing which other modes of communication could be instituted promptly in a manner that preserves parental autonomy [Olusanya, Luxon & Wirz, 2006b; Li et al., 2003]. Parents and health professionals need know that even early introduction of sign language in the early months of life still provides parents of deaf children with a communication mode and has significant benefits for the cognitive and psychosocial development of the child [Schick, Marschark & Spencer, 2006; Meadow-Orlans et al., 2004; Hindley & Parkes, 1999]. Notwithstanding its limitations, sign language offers an opportunity for literacy skills and education and is preferred to the common practice where deaf children are forced to beg for alms as a vocation [Olusanya, Parving & Ruben, 2006]. Early sign language training can be offered to parents and the child in the various schools for the deaf in the country. Presently, there are special schools for the deaf in majority of the 36 states in Nigeria owned/managed by government or missionary organisations. In addition, there are institutions across the country that cater for children with special needs such as hearing loss and blindness. Similarly, intervention to reduce child abuse and neglect can be achieved with better parental education and introduction of legislation against maltreatments. Vocational centres can also be established to expose

children with PCEHL to various trades to enhance their economic independence.

2.4.4. Potential Benefits of UNHS in Nigeria

The primary value of UNHS is the substantial improvement in the age of diagnosis as early as 3 months compared with late detection in the absence of screening [Canale et al, 2006., Harrison, Roush & Wallace, 2003; U.S. Preventive Services Task Force, 2002; Thompson et al., 2001; Davis et al., 1997, Kittrel & Arjmand, 1997]. This early detection of PCEHL ensures that the starting point of any chosen intervention is not from a position of a developmental deficit and also provides a range of benefits which are highlighted below:

2.4.4.1. Prospects for Optimal Language Development

Early detection encourages timely intervention with amplification devices for optimal speech and language development. This is perhaps the most significant advantage of UNHS in Nigeria bearing in mind that majority of children with PCEHL have hearing parents who are less likely to embrace non-verbal communication as their first option [Olusanya, Luxon & Wirz 2005a]. This should vastly enhance the educational and vocational prospects of children with PCEHL in Nigeria and release them from the vicious cycle of ignorance, disease and poverty [Olusanya, Ruben & Parving 2006]. This benefit can only be realised as services for the provision of hearing aids and other associated services become available. Presently, only a few centres offer these services and they are concentrated in the major cities. These centres predominantly serve the needs of adult patients and are rarely tasked with catering for infants because of the lack of early hearing detection services. Where intervention with amplification devices and the associated support services are not feasible, other communication options such as sign language could be promptly initiated for the parent and the affected infant as early as practicable to enhance early childhood development. It may also be the case that many children with profound hearing loss would require

cochlear implants which are currently not offered in Nigeria. Such children may be enrolled promptly for sign language training and avoid the distractions of unorthodox and potentially harmful traditional therapies [Andrade & Ross, 2005; Byford & Veenstra, 2004; Kiyaga & Moores, 2003; Lasisi & Ajuwon, 2002; Odebiyi & Togonu-Bickersteth, 1987].

2.4.4.2. Empowerment of Parents of Hearing Impaired Children

Since the impact of hearing loss extends beyond speech and language development in early childhood its early detection is beneficial to other domains important for optimal early childhood development. For instance, early diagnosis of PCEHL from UNHS in particular empowers parents to relate to their hearing impaired children with better understanding and to seek appropriate and timely help for them. Studies among parents of children with and without hearing loss suggest clearly that the majority of parents appreciate the knowledge of the hearing status of their children as early as possible and therefore consider hearing screening as desirable for their babies [Hergils & Hergils, 2000; Stuart, Moretz & Yang, 2000; Luterman & Kurtzer-White, 1999; Davis et al., 1997; Watkin, Beckman & Baldwin, 1995]. This awareness confers on the parents the right to make informed choices without prejudice to their economic status. This empowerment extends to helping parents to provide the love, nurture and security needed by their child early and is not diminished even where parental denial/adjustment is unduly prolonged [Meadow-Orlans et al., 2004]. Improved parental awareness and knowledge of the value of early detection and intervention is essential to effective parental participation in supporting the affected child which in turn has been demonstrated to have significant positive impact on intervention outcomes [Waktin et al., 2007; Spencer & Marschark, 2006; Moeller, 2000].

2.4.4.3. Growth and Development of Audiological Services

Parents of children with PCEHL that are detected early would naturally desire to help these children as soon as possible. They are not likely to wait till school age before seeking intervention for their children in the schools for

children that are deaf, which they may be forced to do when detection is late. The desire to act after confirmation of hearing impairment could stimulate the development of essential and appropriate intervention services as has been demonstrated in some developed countries with mandatory screening programmes [Uus et al., 2005; White, 2003; JCIH, 2000; Davis et al., 1997]. This in turn should encourage governmental and non-governmental involvement in the management of hearing impairment in newborns, thus increasing the prospect for reforming existing system of rehabilitation of children with profound deafness, which is predominantly limited to sign language.

To facilitate greater access to amplification devices in developing countries, WHO has published comprehensive guidelines for the manufacture of affordable hearing aids, provision of audiological services and training of personnel to encourage auditory-verbal intervention [WHO, 2004a] while low-cost and solar-powered hearing aids are currently produced in Africa [Parving & Christensen, 2004]. Notable charitable organisations like Christoffel-Blidenmission (CBM), Lions Clubs International and Rotary International already have networks for supporting individuals with hearing impairment in Nigeria which can also be channelled towards early hearing detection services in partnership with local non-governmental organizations as well as the National Ear Care Centre (NECC). Already, NECC in partnership with CBM and with technical support from WHO has undertaken a number of initiatives towards implementing WHO guidelines for hearing aids and services in Nigeria.

2.4.4.4. Improved Integration and Educational Support for Hearing Impaired Children

The cultural and social stigma commonly attached to childhood disabilities in general leads to isolation and neglect of essential support services for children who are deaf or hard of hearing. The lack of legislative provisions for children with disabilities similar to the *Disability Discrimination Act 2005* in the UK or the *Individuals with Disabilities Education Act (IDEA) of 2004* in USA,

tend to foster parental frustration and subsequent maltreatment of the child. Such a provision is a potential spin-off from early hearing detection in Nigeria that would facilitate investment in vital support services for children with hearing loss within and outside the mainstream educational system. The Nigeria's National Policy on Education [Nigerian National Research and Development Council, 1998] seeks to "provide adequate educational opportunities for all learners with special needs to enable them to become independent and contributing citizens" and considers inclusion as the "most realistic form of special education". However, little evidence exist that this provision is currently being implemented for children with hearing loss despite the availability of various practical options. For instance, children with mild to moderate hearing loss can be better supported at school age to fit into mainstream schools through say preferential seating arrangement and better appreciation of their special educational needs by teachers. Those with severe-to-profound hearing loss (and their parents) can be enrolled more promptly into sign-language classes where effective support for the development of spoken language are not immediately available, which is currently not the case in Nigeria. This process may help in generating a more positive cultural change towards hearing-impaired persons and minimize maltreatments and neglect often associated with PCEHL in early childhood [Hibbard et al., 2007; Knutson, Johnson & Sullivan, 2004; Kvam, 2004; Togonu-Bickersteth & Odebiyi, 1985].

2.4.4.5. Possible Gateway to other Newborn Screening Programmes

In some developed countries like UK and USA, newborn screening is routinely offered for congenital anomalies such as phenylketonuria (PKU), hypothyroidism, sickle cell disorders and cystic fibrosis. Although the necessity for such screening programmes have been established for developing countries [Christianson, Howson & Modell, 2006; Howse, Howson & Katz, 2005; Bale, Stoll & Lucas, 2003; Penchaszadeh, 2002], they are still rarely offered. Hospital-based universal newborn hearing screening may provide a possible take-off point, particularly for sickle cell disease in a

country like Nigeria where the condition is highly prevalent [Mgbor & Emodi, 2004; Odetoyinbo & Adekile, 1987; Akinyanju, 1989].

2.4.4.6. Compilation of Relevant Epidemiological Data

Essential data on the prevalence and pattern of PCEHL is difficult to obtain without UNHS. Current data are extrapolated from studies in developed countries or from studies among infants and young children without aetiological diagnosis of PCEHL. Risk factors for PCEHL may vary across communities and UNHS is helpful in identifying, measuring and tracking them. The data gathered can be used in planning services for the efficient management of PCEHL. Such data are also needed to justify resource allocation for systematic capacity-building within the healthcare system for hearing impairment [Davis et al., 1997]. In addition, UNHS provides a unique opportunity to obtain other essential data on maternal and child care related to stillbirths, maternal and neonatal mortality patterns.

2.4.5. Risks Associated with Infant Hearing Screening

Although UNHS has been widely accepted as an essential component of an ideal public health service, a number of limitations are still associated with the screening of all babies before hospital discharge, typically during the first 48 hours after birth.

2.4.5.1. False Assurance from Late-Onset PCEHL

False negative results from cases of late-onset and acquired hearing impairment. This has significant implications for developing countries where significant permanent childhood hearing impairment is postnatally acquired from sepsis, neonatal jaundice, measles, mumps, meningitis and the commonly used ototoxic drugs such as chloroquine and gentamicin. There is in fact no one-time or single screening protocol that can detect all categories of hearing loss "early enough". Consequently, on-going surveillance is crucial and must be added for timely detection [JCIH, 2000].

2.4.5.2. False Assurance from False Negative Outcomes

Closely related to this first issue is the false assurance that mothers may receive following a false-negative test because their babies have delayed or progressive permanent hearing loss. For instance, asymptomatic CMV infection may account for significant hearing loss in children lacking risk factors at birth [Barbi et al., 2003; Williamson et al., 1992; Fowler et al., 1999]. The NICU population is especially at higher risk for these categories of sensorineural hearing loss from CMV infection, severe respiratory failure, persistent pulmonary hypertension and mechanical ventilation [Williamson et al., 1992; Fowler et al., 1999; Hutchin, Gilmer & Yarbrough, 2000; Mann & Adams, 1998; Robertson et al., 2002].

2.4.5.3. False Assurance from Undetected Minimal Hearing Loss

Furthermore, most screening protocols do not detect all mild PCEHL and some screening programmes do not make provision for children with unilateral PCEHL of any severity [Johnson et al., 2005]. These limitations are of concern because of the impact of these categories of hearing impairment on the cognitive, psycho-social and educational development of the affected children [Downs, 2007; Teasdale & Sorensen, 2007; Bess, Dodd-Murphy & Parker, 1998; Davis et al., 1986]. On-going surveillance and periodic hearing tests in early childhood are valuable to address this problem [JCIH, 2000].

2.4.5.4. The Burden of False Positive Outcomes

The possible high incidence of false positives resulting from outer ear obstructions may constitute a major burden on the existing healthcare system. This may make the screening programme to be more expensive and ultimately limit its coverage due to budget constraints. Similarly, false positives are associated with undue parental anxiety. However, this concern may be partly addressed by delaying screening, as far as practicable, till 24-48 hours after delivery [Davis el., 1997]. Also, the use of AABR as the second stage screen after TEOAE may reduce false positive rate by almost 50% [Kennedy et al., 2000].

2.4.5.5. Effects on Parent-Child Bonding

The early knowledge of hearing loss in a newborn may be associated with adverse effects on parent-child bonding. A number of parents have experienced some difficulty in this area [Fitzpatrick et al., 2007; Stuart, Moretz & Yang, 2000; Paradise, 1999]. However, only few parents often expressed such concerns and they, in fact, would still prefer, along with the vast majority of parents, to know early than later [Fitzpatrick et al., 2007; Young & Tattersall, 2007; Meadow-Orlans et al., 2004].

Evidence from various on-going UNHS worldwide strongly suggests that parents consider that the benefits of screening far outweigh the associated risks [Olusanya et al., 2007; Bamford, Uus & Davis, 2005; White, 2003; 2004; Stuart, Moretz & Yang, 2000; Watkin, Beckman & Baldwin, 1995]. In fact, parents are displeased when their babies are not offered hearing screening services or when they are ahead of healthcare professionals in detecting developmental problems [Russ et al., 2004; Glover, 2003; Magnuson & Hergils, 2000; Luterman & Kurtzer-White, 1999; David et al, 1997]. Besides, many parents have painful feelings and great sense of guilt for missing the opportunity to know early that their child had some detectable developmental problems [Mehl, 1999; Glover, 2003].

Given the current experiences with late detection and sub-optimal intervention with late enrolment in the schools for children that are deaf; the prospects for language development and better family-integration through auditory-verbal intervention are likely to have an overriding consideration for parents. Even in the absence of auditory-verbal intervention, the early detection of PCEHL offers considerable benefits over the potential risks often associated with traditional and unorthodox therapies or the search for "spiritual cure" for deafness which parents in Nigeria may be inclined to try out of frustration and ignorance [Stephens, Stephens & Eisenhart-Rothe, 2000; Lasisi & Ajuwon, 2002; Odebiyi & Togonu-Bickersteth, 1987].

2.4.6. Potential Challenges for EHDI in Nigeria

Besides the concerns commonly associated with infant hearing screening described in the preceding section, the introduction of infant hearing screening programme in Nigeria is likely to be confronted with the following challenges peculiar to developing countries.

2.4.6.1. Manpower Shortages for Audiological Services

Acute shortage of ear-care professionals is a peculiar feature in Nigeria as in many other developing countries [Eleweke, 1997]. The significant strides in the fields of audiology and audiological medicine witnessed in the last two decades in the developed world are yet to extend to developing countries [Luxon & Barrenas, 2000]. For instance, otolaryngologists per million children under 15 are as high as 320 in Europe compared to one otolaryngologist to 1.2 million Nigerians [Alberti, 1999; Chukwuezi, 2000]. Formal full-time training for audiology and speech pathology are lacking in most tertiary institutions. The only available training is offered in two universities as part-time and post-graduate diplomas for candidates in special education. The average period spent in Ear, Nose and Throat or Audiology posting by medical students is less than 4 weeks during their undergraduate training. The diagnostic capabilities of most tertiary hospitals are limited to pure tone audiometry and tympanometry. Hence, objective diagnostic tests such as OAE and ABR are not readily available [Okeowo, 2001].

However, it is envisaged that, the recent establishment of a National Ear Care Centre (NECC) by the Federal Ministry of Health, will help in addressing the manpower needs in this specialty. NECC has already commenced the training of middle level manpower in ENT as well as primary ear care technicians using WHO guidelines on capacity-building for hearing aids and services, and the Primary Ear & Hearing Care Training Resource [WHO, 2006a & 2004a]. The skills requirement for infant hearing screening is minimal and this function along with some basic rehabilitation services can

be readily handled by non-specialists [Wirz & Lichtig, 1998]. In addition, the new provisions for early hearing detection in the Revised National Health Policy for Nigeria should enhance the prospects for equipping public hospitals at the tertiary and secondary levels to deliver diagnostic and rehabilitation services.

2.4.6.2. Scarcity and High Cost of Ear Care Services

Availability and cost of amplification devices and related services are also of concern. These services are currently not available in many public hospitals but in private centres at prices that may not be readily affordable. However, WHO in partnership with various non-governmental organisations such as Christofell-Blindenmission, World Wide Hearing and Lions Clubs International are working towards the provision of functional, easy to maintain and affordable hearing aids and services for developing countries [WHO, 2006a & 2004a; Parving & Christensen, 2004]. Unlike developed countries ear care services may not be provided free as with most other health services due to limitations in public funding. In fact, out-of-pocket spending still accounts for a significant proportion of total health expenditure in Nigeria. It is therefore possible for infant hearing screening to start as a non-governmental initiative with minimal public funding as experienced in a growing number of developing countries [Olusanya et al., 2007]. The newly introduced national health insurance scheme offers an additional prospect for making these services more widely available.

2.4.6.3. Poor Awareness among Health Workers

Physicians' awareness and attitude towards childhood hearing impairment is a critical factor for success for UNHS. Not all doctors and other health workers can be presumed to be up-to-date in their knowledge of advances in otology/audiological medicine and the range of possibilities currently available for the hearing impaired infants. The health professionals are heavily relied upon for opinion on medical conditions and they wield considerable influence on parents who may be in denial or are simply reluctant to accept prescribed intervention.

Awareness workshops for health professionals especially at birthing centres and prospective infant hearing screening locations are valuable in providing relevant up-to-date information while current medical curricula should make provision for improving the knowledge of medical students [Lock, 2003]. The involvement of nurses and midwives in this educational process is equally essential.

2.4.6.4. Difficulties with Tracking and Follow-up of Parents

Some parents of babies who fail at any of the screening stages may not actively take part in follow-up for several reasons such as: denial, desire to seek non-medical intervention or unfavourable socio-cultural beliefs on hearing impairment including the use of hearing aids [Swanepoel, Hugo & Louw, 2006; Andrade & Ross, 2005; Kiyaga & Moores, 2003; Stephens, Stephens & Eisenhart-Rothe, 2000; Odebiyi & Togonu-Bickersteth, 1987]. The task of completing the screening process through to diagnosis and appropriate/timely intervention may be difficult due to geographical location and socio-economic circumstances of the parents especially in relation to transportation costs [Mukari, Tan & Abdullah, 2006; Isaacson, 2000]. Some parents may simply not be interested in continuing the screening after the initial failed screen even without any physical or financial constraints because the evidence of PCEHL is not yet apparent or due to wrong advice from some health professionals [Finitzo, Albright & O'Neal, 1998]. Poor follow-up rate can also be attributed to administrative lapses and lack of an efficient tracking system [Mukari, Tan & Abdullah, 2006]. Default rates could be minimised with properly administered tracking system, better parental education, and personal contacts with parents including securing accurate contact details and efforts to subsidise or eliminate transportation costs for follow-up visits [Isaacson, 2000].

These challenges may appear daunting but are not insurmountable as demonstrated by the growing number of infant hearing screening programmes in other developing countries in Latin America, Middle East, and East Asia and Pacific through the systematic and dedicated engagement of relevant stakeholders in on-going advocacy and public-private partnerships [Olusanya et al., 2007].

Chapter 3

Materials and Methods

3 Materials and Methods

3.1 Research Design

This research was designed as a cross-sectional, prospective, community-based study over a period of 40 weeks for a population of newborns in an inner-city maternity hospital and for infants attending four BCG immunisation clinics in Lagos, Nigeria within the context of primary health care service.

3.2. Outcome Measures

3.2.1. Performance Criteria for Screening Models

The effectiveness of early detection is best measured by the improvement in outcomes in the domains of language, cognitive and behavioural as well as educational achievement. Such an evaluation often requires a long-term follow-up of children with PCEHL up till school age. However, the NIH [1993], JCIH [2000] and NHSP, UK (www.nhsp.info) have outlined quality indicators that are commonly regarded as "intermediate" outcomes or surrogates of the long-term outcomes for evaluating the effectiveness of infant hearing screening programmes [Watkin, 2003; Davis et al., 1997]. These indicators have been adopted for this study and principally consist of the following variables:

I. Screening coverage – as measured by the percentage of infants screened before hospital discharge or within three months of age among those eligible for screening [NIH, 1993].

II. Screening effectiveness - as measured by the referral rate for diagnostic evaluation following the two-stage screening process among those completing the protocol.

III. Return rate for diagnostic evaluation.

IV. Age of confirmation of hearing loss.

V. Effectiveness of screening protocol – based on the sensitivity, specificity and the likelihood ratios of the two-stage screening protocol.

3.2.2. Comparative Performance of Screening Models

The comparative analysis of the merits and drawbacks of each of the screening models were based on the following criteria [Vohr et al., 2001; Headley, Campbell & Gravel, 2000; Davis et al., 1997; Maxon et al., 1997]:

1. No. of babies screened per day
2. Referral rates for TEAOE and AABR
3. Analysis of screening cost per infant.
4. Yield for PCEHL

3.2.3. Risk Factors for PCEHL in Nigeria

Risk factors used in this study were primarily derived from the JCIH 1994 [AAP 1995] and JCIH 2000 position statements (Table 3.1). However, risk factors that could not be elicited or established in our target population were excluded while those that were considered relevant in our screening environment but not included in the JCIH lists were incorporated. For instance, mechanical ventilation was not available in the hospital used in this study and was therefore excluded as a potential risk factor.

Consanguinity, not listed by JCIH, has been associated with PCEHL in Nigeria and was therefore included in the list of potential risk factors [Olusanya, Luxon & Wirz, 2005b; Ijaduola, 1982]. Because of the difficulty of identifying or confirming maternal rubella, maternal rash in pregnancy which has been associated with hearing loss in a developing country was used as a proxy [D'Mello, 1995]. A child born by a mother with HIV/AIDS may have a high risk of PCEHL due to increased vulnerability to infections such as cytomegalovirus [Vancikova & Dvorak, 2001; Meynard et al., 1997]. Positive maternal HIV status was therefore added to the list of risk factors.

Table 3-1 Potential Risk Factors for PCEHL in Current Study

Risk Factors
Prenatal
• *Family history of hearing loss.*
• *Consanguinity*
• *In-utero infections such as rubella, herpes, or positive maternal rash in early pregnancy*
• *Maternal HIV*
• *Maternal malaria*
• *Congenital malaria*
• *Craniofacial anomalies*
• *Stigmata or other findings associated with a syndrome known to include a sensorineural and/or conductive hearing loss*
Postnatal
• *Non-hospital delivery*
• *Prematurity <34 weeks*
• *Birth weight less than 1,500g (3.3lbs)*
• *Apgar score ≤ 4 at 1 minute and ≤ 6 at 5 minutes or history of birth asphyxia*
• *Hyperbilirubinaemia at levels requiring exchange blood transfusion*
• *Ototoxic medications.*
• *SCBU/ hospital admission in the first 28 days of life*
• *Bacterial meningitis.*
• *Small-for-gestational-age (SGA)*

A high proportion of births without skilled birth attendants have been recorded in Nigeria [WHO, 2006b; UNICEF, 2006]. WHO defines skilled birth attendants as doctors, nurses and healthcare workers with formal training but excludes traditional birth attendants. Several reports have demonstrated adverse obstetric outcomes for mothers who opt for such non-institutional/hospital services. Consequently, non-hospital delivery was added to the list of risk factors used for this study. Similarly, malaria and its treatment with ototoxic drugs such as quinine and intravenous chloroquine may be associated with hearing loss [Shine & Coates, 2005; Hadi, Nuwayhid

& Hasbini, 1996, Chukuezi, 1995, Obiako, 1985]. Hence congenital malaria was listed as a risk factor.

However, congenital malaria (defined as high maternal, placental and cord blood parasitaemia) may sometimes be difficult to diagnose. Therefore, maternal malaria during the last trimester of pregnancy was used as a proxy [Okafor, Oguonu & Onah, 2006; Runsewe-Abiodun, Ogunfowora & Fetuga, 2006; Obiajuwa, Owa & Adeodu, 2005]. Intra-uterine growth retardation/small-for-gestational-age are a significant feature of congenital malaria in the newborn besides haemolytic jaundice, anaemia and hepatosplenomegaly from the heavily plasmodium falciparum parasitized placenta. For these reasons, small-for-gestational-age was added to the list of potential risk factors. Importantly small-for-gestational-age is also a feature of anomalies associated with consanguinity and intrauterine infections [Mumtaz et al., 2007; Boppana et al., 1992].

A history of hospital admission in the first 28 days of life was also included as an index for serious conditions or illness that could not be readily diagnosed but could be associated with PCEHL such as neonatal meningitis and sepsis.

3.3. Study Location and Settings

An important consideration in the choice of locations for this pilot study was availability and proximity of support services for children detected with PCEHL within the programmes. For this reason, metropolitan Lagos within Lagos State in the Southwest of Nigeria was selected out of the 36 state capitals in the country for this study.

Lagos State has an estimated population of about 15 million people and is the most densely populated state in Nigeria with 94 percent of its population urbanised. It is divided into 15 administrative councils or Local Government Areas (LGAs) which are responsible for primary education, collection of vital statistics, sanitation and primary health care including maternal and child health services. The city of Lagos is situated on the Gulf of Guinea (Figure 1.2) and is officially regarded as the second largest city in Nigeria, after Kano in the North. However, unofficial reports indicate that Lagos is perhaps the largest city in sub-Saharan Africa. Although Lagos city ceased to be the Nation's capital in 1991, it remains the economic centre for the country with the largest concentration of industries, financial firms, educational institutions and the nation's chief seaport and busiest airport.

One of the LGAs, known as Lagos Island, with a population of 243,777 was further selected because it was most suited logistically for conducting pilot programmes on infant hearing screening (Figure 3.1). This inner city area, apart from being the most active financial and commercial centre in Lagos and the country, is served by one general hospital, one children's hospital, one maternity hospital, seven health centres (all of which are State-owned), several private hospitals and herbal homes (i.e. traditional maternity homes). An audiological centre that offers comprehensive diagnostic and rehabilitation services is also located in this LGA.

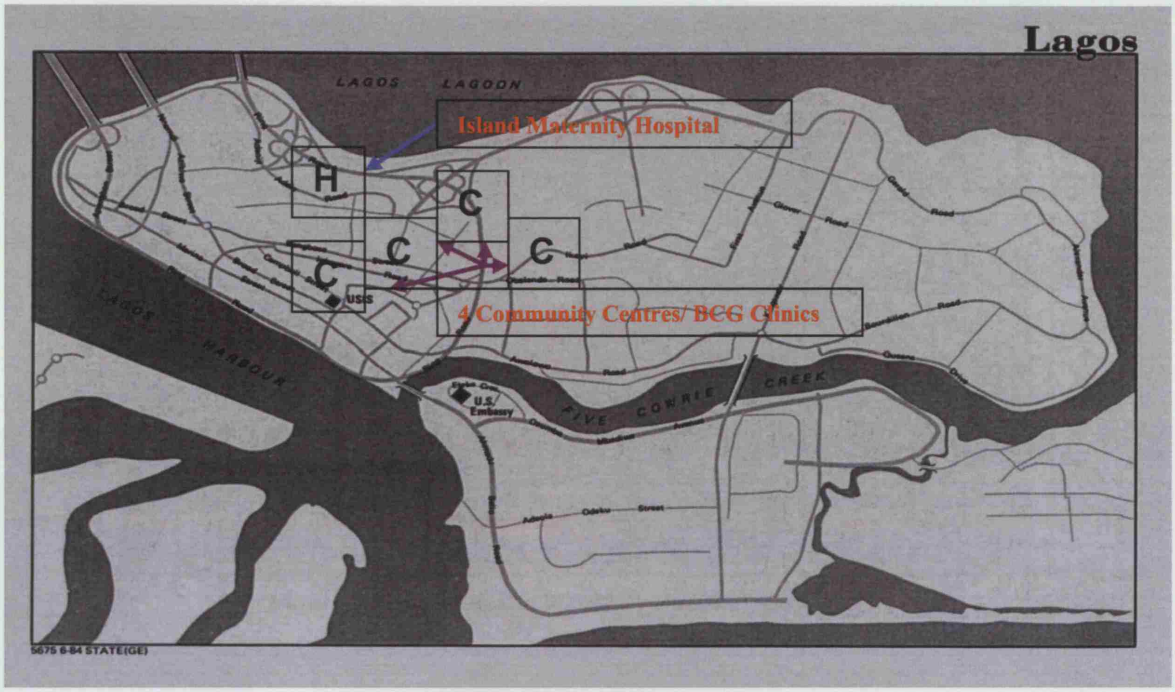


Figure 3.1. Map showing Lagos Island Local Government Area
Key to Screening Sites: H = Hospital; C = Community Health Centre

3.3.1. Choice of Hospital-based Screening Site

Lagos Island Maternity Hospital was chosen for the hospital-based programme because it is the only specialised maternity hospital in the study location and has the largest single annual birth rate of 2,400 babies. Her Royal Highness, the Duchess of Gloucester laid the foundation stone of the hospital in May 1959 and it was formally declared opened in July 1960 by Lady Robertson, wife of the then Governor-General of Nigeria. At the time of this study, the hospital had 180 beds for maternity services distributed over three floors (Figure 3.2). It was managed by the Federal Government as a federal hospital for many years before it was subsequently ceded to the Lagos State Government.

Figure 3.2 Lagos Island Maternity Hospital, Lagos

The hospital serves as a referral centre for over 300 private and public hospitals in the Lagos metropolis and its environs and provides residency training in obstetrics and gynaecology as an accredited institution for National Postgraduate Medical Training in Nigeria. Patients attending the hospital are not required to pay for antenatal care and straightforward delivery. However, an equivalent of about £20 (Naira 5, 300 in local currency) is required for blood tests and delivery pack. Drugs and special procedures such as caesarean operation attract extra charges.

3.3.2. Choice of Community-based Screening Sites

Routine childhood vaccinations are widely administered across community or primary health centres in Nigeria under the auspices of a Federal Agency on the National Programme for Immunisation (NPI). A review of the uptakes for various childhood vaccinations prior to the commencement of the study showed that BCG immunisation was highest and consistent with the annual data from UNICEF [2006]. BCG immunisation is given shortly from birth and is the earliest vaccination for children in Nigeria. This clinic was therefore selected for the purpose of the screening programme. The community-based screening site was required to be in close proximity with the hospital-based programme for better coordination by the researcher. Routine immunisation

clinics in Lagos Island LGA were held on different week days from Monday to Thursday in seven primary health care centres within the vicinity of the hospital-based hearing screening programme. This meant that there were some days when clinics were held in more than one location.

At the onset of the screening programme, all babies attending the various immunisation clinics were referred daily after receiving the BCG vaccination to a central location for hearing screening. However, the uptake remained very low at the end of the first two months despite regular educational campaigns to parents when attending the clinics. Investigation by the researcher revealed that parents were not willing to bear the cost and inconvenience of a separate visit to a clinic solely for a newly introduced programme for a "non-life threatening condition". To reverse this trend, screening was promptly re-arranged to take place on-site at the immunisation clinics from Monday to Thursday. Since there was only one screening team, it was necessary to select four sites to be enlisted for the programme (Figure 3.3). These primary health centres were Lagos City Hall Health Centre, Massey Street Health Centre, Onikan Health Centre and Sura Market Health Centre.

These four sites were located within a radius of about 2 kilometres and provided the best possible combination of screening locations accounting for 75.1% of the total attendance of 2,773 babies who received BCG vaccination in the preceding six months (January – June 2005).

Lagos City Hall Community Health Centre

Massey Street Community Health Clinic

Onikan Community Health Centre

Sura Market Primary Health Care Centre

Figure 3.3. Sites for the Community-based Screening Programme

3.4. Ethical Clearance and Institutional Support

Ethical approvals were obtained from the University College London, UK (Appendix 3.1) and the Lagos State Health Management Board, Nigeria (Appendix 3.2). The local ethical approval was granted on the condition that parents would not be required to pay for any of the services to be provided under the research project including the provision of hearing aids. An official letter of introduction to all heads of public hospitals in Lagos Island LGA was obtained by the researcher from the State Health Management Board. This was to give the researcher freedom to choose the most appropriate hospital for the study. Against the backdrop of the prevailing low public perception of childhood hearing loss as a non-life threatening condition, institutional support at the highest possible levels were considered valuable to ensure good participation from all levels of health workers and parents. Accordingly,

the researcher sought and secured the official launching of the hospital-based programme shortly after its commencement by the Federal Health Minister accompanied by the Lagos State Health Commissioner and other high ranking health officials.

3.5. Research Participants

3.5.1. Hospital-based Programme

All babies born in the hospital from the commencement day of 16th May, 2005 to February 28, 2006 were eligible for enrolment into the study. Babies born before 16th May, 2005 and awaiting discharge were also enrolled in the study.

3.5.2. Community-based Programme

Although BCG vaccination was meant to be given at birth or shortly thereafter, some parents did not attend the clinic until their babies were older sometimes up to 12 months of age. Since one of the quality indicators for this project required babies to be screened within the first three months of life, infants older than 3 months at the time of attending the BCG immunisation clinics were excluded. This exclusion also became necessary because older babies are often restless and easily irritable and thereby difficult to test without sedation, which was considered an unnecessary burden for the screening staff. Moreover, infants older than 3 months were more prone to false positives because of persistent otitis media with effusion [Boone, Bower & Martin, 2005; American Academy of Family Physicians (AAFP) et al., 2004; Bantock & Croxson, 1998, Paradise et al., 1997]. All mothers attending the clinics were notified of this criterion during the routine pre-vaccination briefing by the community nurses. The screening team also verified the ages of prospective participants before screening was offered.

3.6. Research Team

3.6.1. Programme Coordinator

The screening programmes were directly supervised by the researcher. The researcher visited successful UNHS programmes in Phoenix, USA and in London in preparation for this research project. A one-day personalised training session was specially arranged in London for the researcher by the National Training Coordinator for the NHSP (UK). Demonstration sessions at hospital locations in London, Phoenix and Ghana were also arranged for the researcher by the manufacturers of the screening equipment. The researcher also brought to bear on this project over 8 years clinical experience in electrophysiological measurements of the peripheral auditory system in a developing country setting.

3.6.2. Screening Staff

3.6.2.1. Selection Criteria and Duties

Two separate teams of screeners were recruited, one each for the hospital- and community-based programmes. Each team consisted of two non-specialist staff, based on the experiences from UK and other countries with successful on-going UNHS programmes and the need to demonstrate the practicability of infant hearing screening in a primary care setting. One of the staff had prior experience as an auxiliary nurse (nurse assistant or health worker) in private hospitals and the second member of staff had basic secondary education with no prior experience in healthcare. The community-based team had an additional support staff to assist in managing the crowd in very busy clinics. Individual staff selection was based on good references from persons known to the researcher. Good interpersonal skills, shared vision of the screening programme, and willingness to learn and participate as a member of the screening team in a hospital or community setting were principal considerations for recruitment. The screeners were competitively remunerated to foster a high sense of devotion to duty and forestall avoidable

staff turnover from the generous support received from a local non-governmental organisation.

The support staff was responsible for obtaining written parental consent and completing the patient data entry forms while the auxiliary nurse was responsible for conducting the hearing test and anthropometric measurements for the babies.

3.6.2.2. Training Curriculum and Guidelines

Training is essential for non-specialist with no prior audiological experience to effectively conduct an infant hearing screening programme particularly in a primary care setting. The training objective therefore was aimed at equipping the screening team with the knowledge, skill and attitude to confidently and successfully implement a hospital- or community-based UNHS programme. The training was interactive and was personally conducted by the researcher. The individual knowledge-base was achieved through lectures, group work, assignments, and regular theoretical assessments (Appendix 3.3). The acquisition of relevant skills was based on clinical and video demonstrations as well as role-play. Trial screening under the supervision of this researcher was arranged during the second week of training in the audiological centre that supported this programme.

Practical guidelines covering troubleshooting with screening instruments were drawn up and carefully reviewed with the staff as detailed in Appendix 3.3. By the end of the training programme, participants understood the concept of early detection, the limitations of a screening programme and the importance of follow-up. In addition they demonstrated readiness to take ownership of the programme and were also confident enough to communicate with parents in the hospital or community health centre. The screening staff were under strict instruction to contact the researcher promptly whenever they encountered any difficulty with their tasks with respect to the research project or to contact the appropriate hospital staff in a

situation that involved parental query or concern about other child healthcare issues.

3.6.2.3. Duration of Training

The training was held outside the screening sites for a period of two weeks. Although it has been suggested that a 2 – 4 hours training in the use of the screening instruments was adequate [Maxon et al., 1997], it was necessary to consider the special needs and possible challenges of introducing such a new programme in a developing country. Hands-on training under the direct supervision of the researcher continued in the first four weeks of the hospital-based programme for the two teams. Thereafter, the second team moved into the community and were directly supervised also by the researcher for the first 4 weeks. The two weeks of training and the direct supervision for the first four weeks of screening were more than adequate in helping anyone without prior audiological experience to handle the fully automated and simple-to-use screening instruments confidently and effectively. Notwithstanding the researcher made several unscheduled visits to the screening sites during the study period as part of the quality control initiatives; or when required to resolve matters involving the hospital staff or community health workers.

3.6.3. Nurse Educators

An awareness workshop on the significance and purpose of the screening project was briefly conducted by the researcher for doctors and nursing staff at Island Maternity Hospital on the commencement day of screening, during which the screening teams were also introduced. As part of the educational programme an information booklet [www.soundstart4all.com] addressing the following 10 important issues was distributed to health professionals and parents:

1. What happens when a child is unable to hear?
2. Is hearing loss a common problem in Nigerian children?
3. What are the main causes of childhood hearing loss?

4. Are the risk factors for hearing loss preventable?
5. What can be done when primary prevention fails?
6. What does the screening test entail?
7. What happens when the screening results are unsatisfactory?
8. How can the government help parents?
9. What role can the public play to support government?
10. What is the healthcare worker's role?

The assistance of the Sisters-on-Duty was solicited in educating parents on the programme during ante-natal clinics and as soon as they were admitted or shortly thereafter as part of the routine hospital briefing for new patients. The entire hospital staff were enthusiastic about the screening programme especially because it was to be offered at no charge to the parents. The knowledge that newborn screening was a standard of neonatal care in developed countries and that the hospital was the first in the country to run such a programme, as well as the subsequent official launch by the Federal Health Minister, provided additional motivation for the hospital staff and parents. A similar introduction was provided for the community nurses in charge of the immunisation clinics at the four community health centres.

3.6.4. Data Entry Clerk

A data entry clerk with good computer skills was engaged on a part-time basis to assist the researcher with transferring the information collected at the screening sites into the data management software programme. Only information duly verified by the researcher was made available to this staff member and as such he had no direct link with the screening teams. He promptly notified the researcher when errors were spotted in the records at the point of data entry or for any other related queries.

3.7. Test Environment

The conditions of the environment in which the screening instruments were used were important factors that were considered before the commencement of screening. For instance, the adverse effects of excessive ambient noise on TEOAE recordings such as prolonged testing time, the inability to obtain

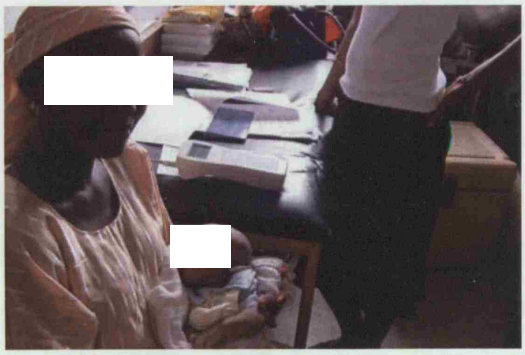
accurate recordings and high false-positive rates have been well documented [Headley, Campbell & Gravel, 2000; Rhoades et al., 1998; Hunter et al., 1994, Jacobson & Jacobson, 1994]. In contrast, AABR tests are less susceptible to background noise. However, they are difficult to conduct when there is myogenic interference or when the baby to be tested is restless and irritable thereby prolonging the testing time. Consequently, the ambient noise levels were measured with a Larson Davis Type I Precision 800 dB Integrated Sound Level Meter, as part of the trial tests with the TEOAE instruments in each of the screening locations to determine the most suitable sites for conducting the tests.

3.8. Screening Instruments

3.8.1. Echo-Screen TEOAE Screener

Two models of automated TEOAE screeners – Echocheck donated by Otodynamics UK and Echo-Screen (Figure 3.4) loaned from Natus Medical Inc, USA - were available for this project.

(a) TEOAE Screening at the Hospital



(b) TEOAE Screening at the Community Centre

Figure 3.4. TEOAE Screening

The 'Noise OK' indicator of the Echocheck designed to monitor the ambient noise level within acceptable limits often failed to activate even in the quietest sites for screening. Where recordings were possible with the Echocheck, they were more susceptible to high false-referral rates. In contrast, the Echo-

Screen has an in-built noise and artefact rejection system and a summary display of the test conditions at the end of each test. When stimulus stability was less than 80% and more than 20% artefact rate, which may be indicative of a restless infant or excessive ambient noise level, a re-test becomes necessary when the test conditions have improved.

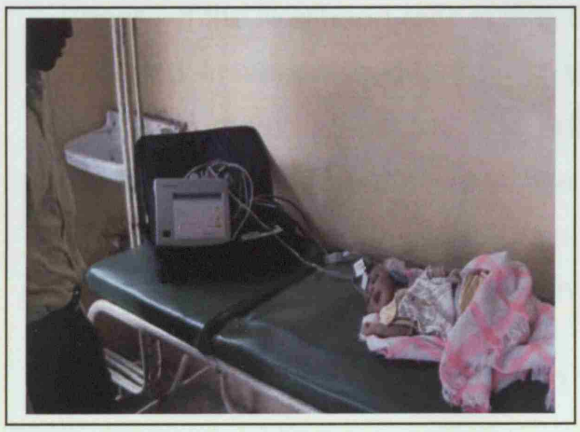
The Echo-Screen has been validated in various newborn hearing screening programmes [Delb et al., 2004; Meier et al., 2004; Grandori et al., 2002] and when powered on, it initiates a routine self-calibration before recordings are made. Transient sounds consisting of 60 nonlinear clicks are presented through a small probe tip in the baby's ear canal. Emissions elicited from the outer hair cells in response to the clicks are picked up by the internal microphone of the equipment. In the default settings, the clicks are presented in a non-linear mode at a peak level of 85 dB SPL. The instrument samples 60 different points within a post-stimulus interval between 6 and 12 ms and then automatically determines the "pass"/"fail" result based on preset binomial statistical probability that an emission has been recorded within the frequency range 1.5 to 3.5 kHz. The tester cannot alter the default settings but could decide to repeat the recording on the basis of qualitative information on the stimulus stability and artefact given by the instrument. A typical recording takes an average of about 2 minutes. The instrument is powered by an-inbuilt rechargeable battery that can last up to 10 hours of continuous use. An important quality control feature of this automated instrument is that every test is assigned a unique identification number which cannot be altered or deleted by the screener until it is downloaded to the central database after a maximum of 250 tests. This feature therefore provides for an independent validation of results recorded by the screeners for each patient.

3.8.2. ALGO Portable AABR Screener

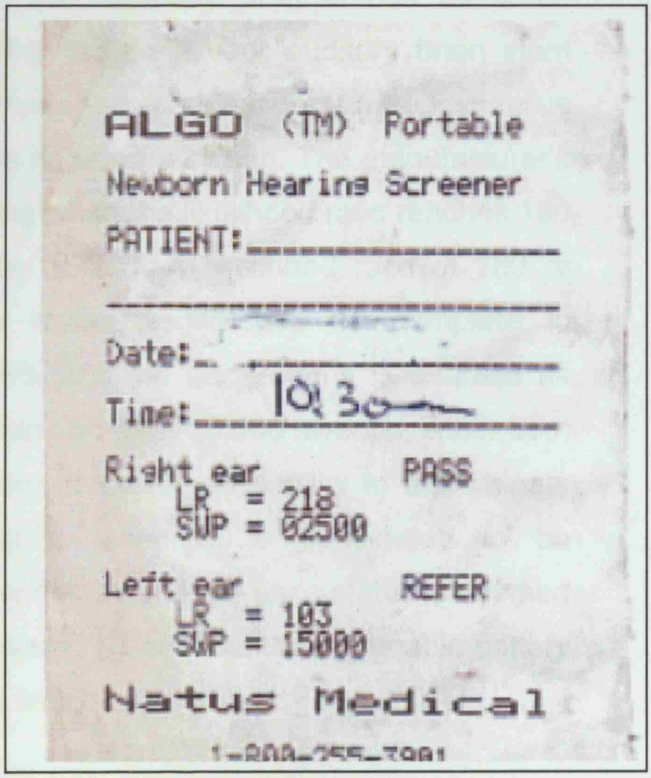
The ALGO automated ABR screening device (Natus Medical California, USA) is perhaps the most widely used model worldwide. It employs a click stimulus with the following parameters:

- Duration – 100 microseconds;
- Intensity – 35 dB nHL; rate – 37 clicks/second, (nominal)
- Acoustic frequency spectrum – 700 to 5000 Hz (±5 dB)

(a) AABR Screening in the Hospital



(b) AABR Screening in the Community Centre



(c) AABR Patient Result Slip

Figure 3.5. AABR Screening at Study Locations

Recording begins with at least one thousand soft click stimuli sounds being presented at 35 dB nHL to newborn's ears through disposable single-use flexi coupler earphones. The responses to the auditory stimuli are recorded with three surface jelly tab sensors or electrodes placed over the vertex, nape and the shoulder (the cheek was used as an alternative site to the shoulder). The sensors pick up the brain wave response to the click sounds and transmit the signals to the screener (Figure 3.5).

In the 'monaural sequential screen mode' the second ear is automatically screened after the first one. During the screening the ALGO screener measures the probability that the ABR is present within the surrounding noise. A "pass" is displayed when the internally programmed template-matching algorithm measures ongoing brain wave or auditory brain stem response at a minimum of 1000 sweeps. The combination of a click stimulus and the corresponding ABR response is called a sweep. The manufacturer's proprietary algorithm terminates testing when the likelihood ratio reaches 160 or when the sweep number reaches 15,000. A likelihood ratio of 160 or greater indicates that the infant's response matches the template to confidence interval of at least 99.98% and the ear's status is deemed as "pass". If the likelihood ratio is less than 160 after 15,000 sweeps, either from reduced or absent auditory brain stem response or inability to discriminate noise from a response, the infant's response is considered to be undifferentiated from a no-response condition and the ear's status is deemed as "refer". This instrument is also powered by an-inbuilt rechargeable battery that can last up to 10 hours of testing time.

3.8.3. Equipment Calibration and Maintenance

All the screening instruments used for this study were within manufacturers' first calibration and no further calibration was recommended for the duration of the study. The instruments also had in-built self-calibration and maintenance status notification mechanism. All equipment was used as recommended by the manufacturer's user manuals.

3.9. Pre-Screening Procedure

3.9.1. Parental Education and Informed Consent

All parents were informed of the consequences of detecting hearing loss late and the benefits of early hearing detection and intervention with the aid of an information leaflet (Appendix 3.4). In the hospital-based programme, the ward sisters effectively discharged this function because of their vast experience with communicating with and educating mothers. They presented the screening programme as part of the immediate post-delivery examination necessary to ensure the new baby had no detectable hearing abnormality that could later impair normal speech and language development. The limitation of the screening procedure in discriminating a hearing loss due to debris or vernix plug in the ear canals from true hearing loss which might require subsequent follow-up visits for necessary confirmation was also emphasised as well as the importance of completing the screening process [Doyle et al., 2000; Maxon et al., 1997; McNellis & Klein, 1997]. Parents were also informed that all the services to be provided under the programme including the provision of hearing aids were at no charge.

Figure 3.6. Parental Education at a Community Health Centre

Similarly, the community nurses at the immunisation clinics educated parents on the advantages and limitations of the screening programme during routine

pre-vaccination health talks while also emphasising the importance of follow-up appointments if required (Figure 3.6). Thereafter, the screening team approached the mothers for their written consent on a detachable section of the programme information leaflets, which were given to all mothers (Appendix 3.4).

3.9.2. Pre-Screening Questionnaire

A structured questionnaire was administered by a member of the screening team to elicit medical and socio-economic history of the mothers and the birth history of the baby or infant (Appendix 3.5). In addition, general physical examination of all babies including anthropometric measurements such as head circumference, length and weight were obtained and documented before screening was conducted.

3.10. Screening Protocols

3.10.1. Hospital-based Programme

The hospital-based protocol for babies screened before discharge is presented in Figure 3.7. The protocol distinguished babies in the Well Baby Nursery (WBN) from those requiring close monitoring after birth because they were sick or preterm and admitted into the Special Care Baby Unit (SCBU).

3.10.1.1. Protocol for Well Baby Nursery

Well babies were first screened with TEOAE between 24 and 48 hours after delivery in the designated room and this test was repeated immediately if a refer result was recorded. As far as possible, babies were only tested after a feed when they were more likely to be well settled. Babies who failed TEOAE tests were scheduled for a second-stage screen with AABR which was usually conducted before hospital discharge. Babies who were referred in one or both ears were referred for a follow-up screen during the routine six weeks post-natal visit to the hospital. In addition, babies who were unable to complete the screening prior to discharge also had a follow-up appointment

to coincide with the first post-natal hospital appointment. Those failing the AABR screening were referred for full diagnostic/confirmatory evaluation at the audiological centre designated for this project.

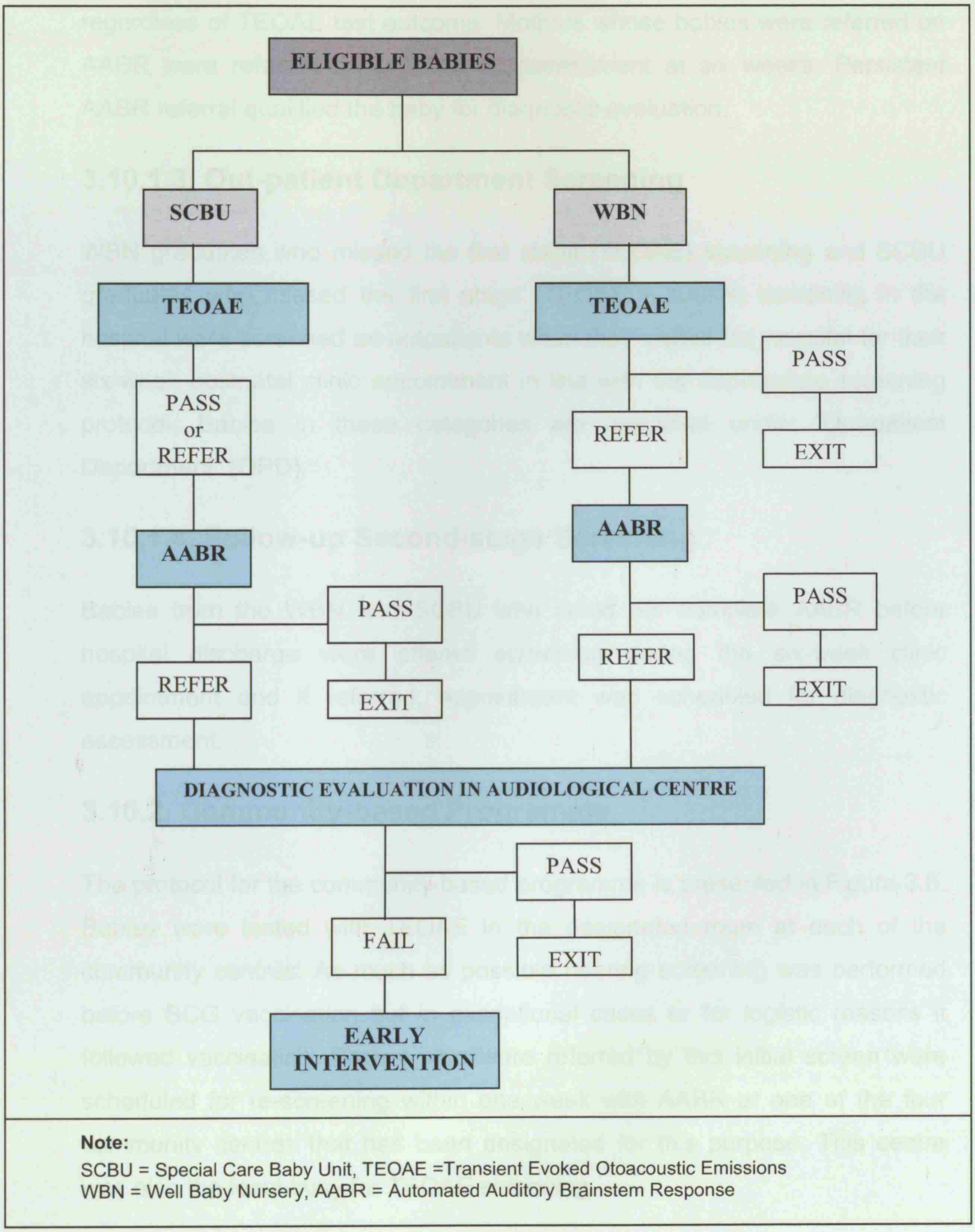


Figure 3.7. Hospital-based Screening Protocol

3.10.1.2. Protocol for Special Care Baby Unit

Babies in SCBU were screened in the designated section of the Newborn Nursery first with TEOAE and then with AABR just before hospital discharge, regardless of TEOAE test outcome. Mothers whose babies were referred on AABR were referred for a follow-up assessment at six weeks. Persistent AABR referral qualified the baby for diagnostic evaluation.

3.10.1.3. Out-patient Department Screening

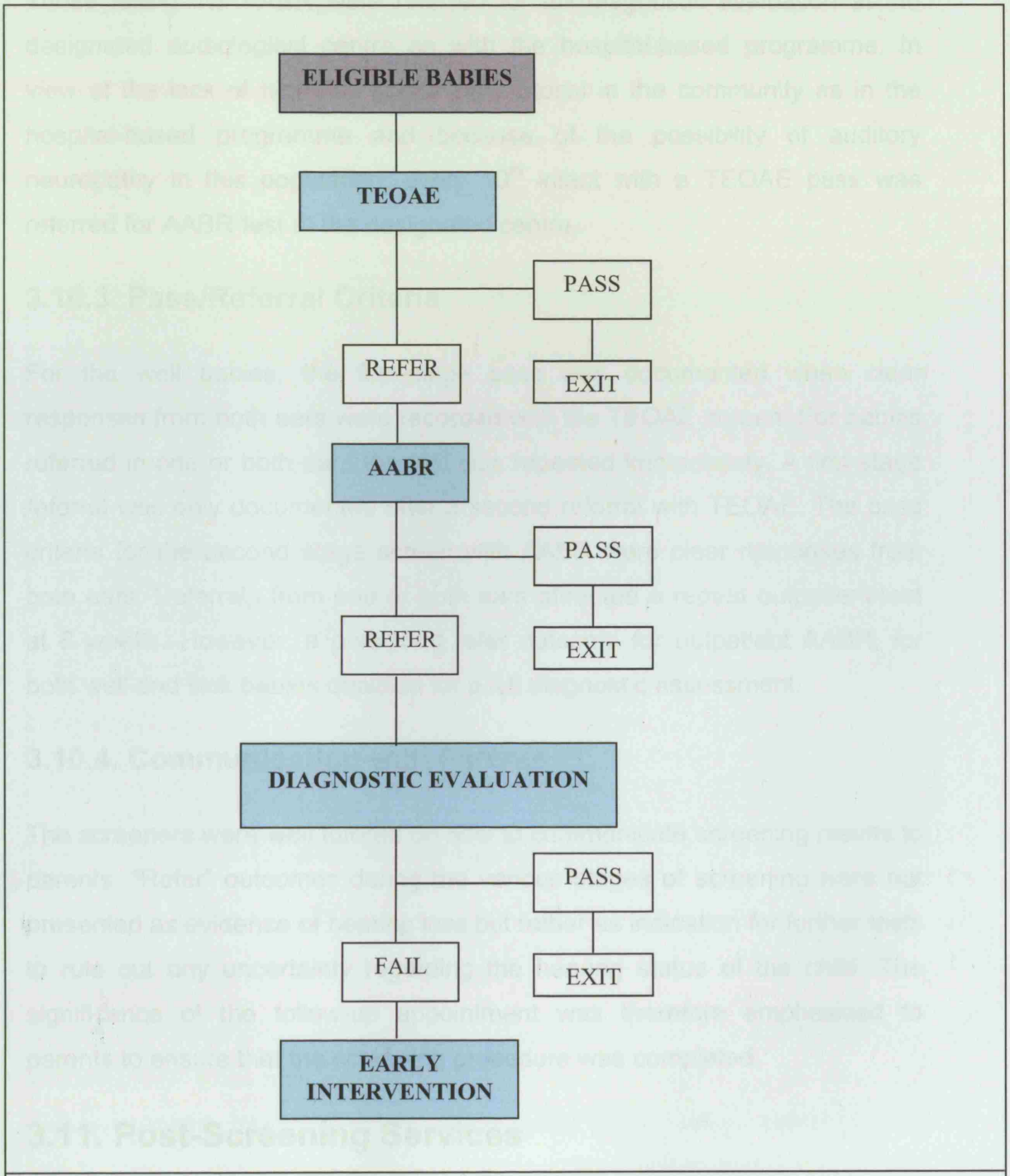
WBN graduates who missed the first stage (TEOAE) screening and SCBU graduates who missed the first stage (TEOAE + AABR) screening in the hospital were screened as outpatients when they visited the hospital for their six-week postnatal clinic appointment in line with the appropriate screening protocol. Babies in these categories are classified under "Out-patient Department" (OPD).

3.10.1.4. Follow-up Second-stage Screening

Babies from the WBN and SCBU who could not complete AABR before hospital discharge were offered screening during the six-week clinic appointment and if referred, appointment was scheduled for diagnostic assessment.

3.10.2. Community-based Programme

The protocol for the community-based programme is presented in Figure 3.8. Babies were tested with TEOAE in the designated room at each of the community centres. As much as possible hearing screening was performed before BCG vaccination but in exceptional cases or for logistic reasons it followed vaccination. Babies who were referred by this initial screen were scheduled for re-screening within one week with AABR at one of the four community centres that has been designated for this purpose. This centre was also the least busy for TEOAE screening.



Note:
TEOAE = Transient Evoked Otoacoustic Emissions. AABR = Automated Auditory Brainstem Response.

Figure 3.8. Community-based Screening Protocol

Those failing the AABR were referred for full diagnostic evaluation at the designated audiological centre as with the hospital-based programme. In view of the lack of high-risk screening protocol in the community as in the hospital-based programme and because of the possibility of auditory neuropathy in this population, every 10th infant with a TEOAE pass was referred for AABR test at the designated centre.

3.10.3. Pass/Referral Criteria

For the well babies, the first-stage pass was documented when clear responses from both ears were recorded with the TEOAE screen. For babies referred in one or both ears the test was repeated immediately. A first-stage referral was only documented after a second referral with TEOAE. The pass criteria for the second stage screen with AABR were clear responses from both ears. Referrals from one or both ears attracted a repeat outpatient-test at 6 weeks. However, a persistent refer outcome for outpatient AABR, for both well and sick babies qualified for a full diagnostic assessment.

3.10.4. Communication with Parents

The screeners were well tutored on how to communicate screening results to parents. "Refer" outcomes during the various stages of screening were not presented as evidence of hearing loss but rather as indication for further tests to rule out any uncertainty regarding the hearing status of the child. The significance of the follow-up appointment was therefore emphasised to parents to ensure that the screening procedure was completed.

3.11. Post-Screening Services

3.11.1. Diagnostic Services

The diagnostic services for this project were provided by an audiological centre designated for this project. It was located within a radius of 4 kilometres from all the screening sites. The centre was established in 1996 and serves as a training and referral centre for teaching hospitals around the

country. Parents were not required to pay for any of these services and as a further incentive to parents, the researcher provided free transportation from the hospital or community health centres where the babies received the screening services to the diagnostic centre.

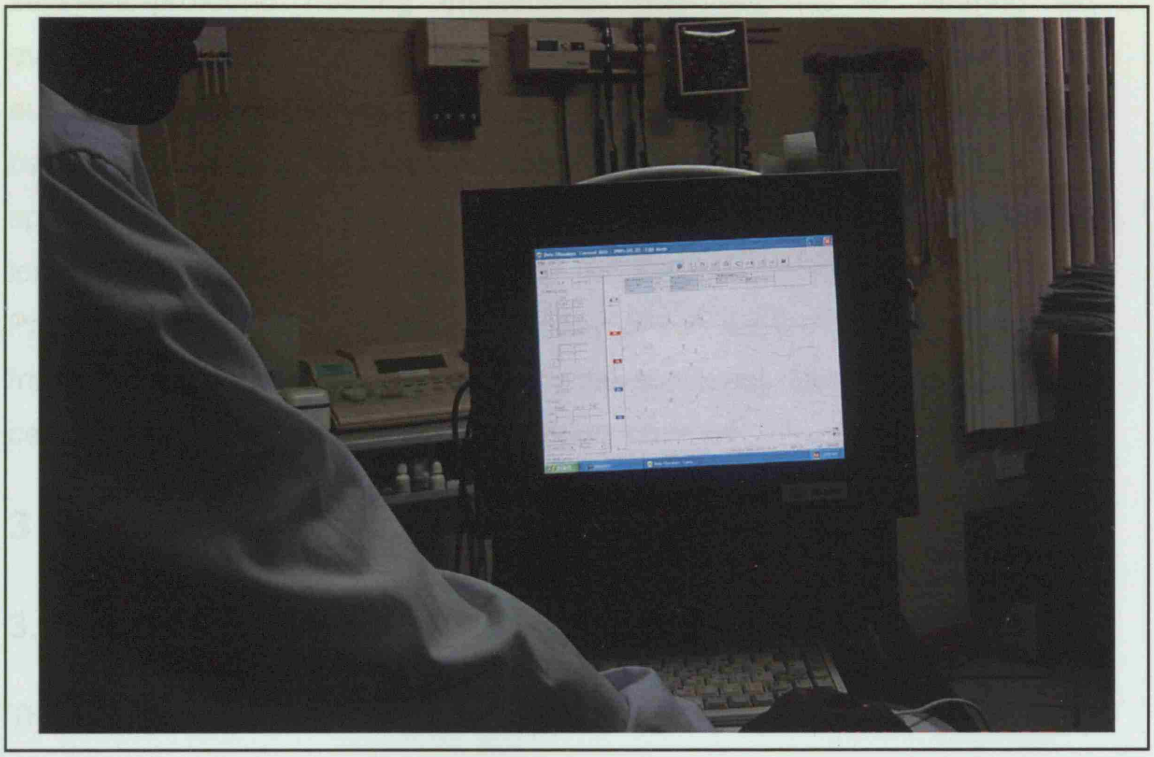


Figure 3.9. Diagnostic ABR [by Interacoustics] at the Diagnostic Centre in Lagos

Diagnostic evaluation for referred babies consisted of tympanometry including high frequency (1000 Hz) probe tone for babies less than 4 months old, diagnostic tone pip ABR with insert ear phones and/or Visual Reinforcement Audiometry (VRA) for babies older than 6 months. The results of the diagnostic assessment which were strictly based on the appropriate parameters and protocols recommended by NHSP (UK) [http://www.nhsp.info/cms.php?folder=21] were available to the researcher. Follow-up counselling appointments were scheduled for the parents of babies who were confirmed with any degree of bilateral or unilateral sensorineural hearing impairment.

3.11.2. Intervention Services

Counselling sessions were arranged by the researcher for parents of all children that failed the diagnostic tests. The purpose was to educate the parents on the implications of the results, the nature of support that would be required for the children and the role expected of parents. Babies with severe-to-profound hearing impairment were referred for hearing aid evaluation and fitting and enrolment into early intervention services at the audiological centre. On-going parental surveillance was recommended for babies with mild-to-moderate or unilateral hearing loss with the aid of a speech and language development chart incorporated into the information leaflet given to all parents prior to the first-stage screening (Appendix 3.4). Parents were not required to pay for any of the intervention services while free transportation was provided from the screening site to the diagnostic centre which also provided the intervention services.

3.12. Data Management

3.12.1. Data Collection

The questionnaire used in this study was designed as the principal data entry form (Appendix 3.5). The screening team at each location was responsible for completing this form for each patient prior to and after the screening tests. The completed forms were delivered to the researcher on a daily basis. The researcher regularly checked each form and notified the screening teams of any queries especially relating to missing information. All duly verified forms were handed over to the data entry clerk. The researcher independently obtained the hospital records for deliveries on a monthly basis to verify the babies who had missed screening as reported by the screening team as part of the quality control for the programme. Similar data for children attending the BCG immunisation clinics were also collected independently by the researcher on a regular basis to confirm that those who were reported to have received the BCG vaccination by the health records but were not screened met our exclusion criteria.

3.12.2. Data Management Software

A major and unique challenge for a population-based infant hearing screening with a large dataset on the subjects' demographics is the tracking of results through the many stages of the programme from the first screening test to enrolment into intervention services. Poor management of these data can significantly compromise the reliability and integrity of the reported outcomes. Consequently, a well tested data tracking and management software – HI*TRACK for Windows Version 3.5 Desktop 04-02-2004 (National Centre for Hearing Assessment and Management: NCHAM, Logan, UT, USA) – was used in this study for monitoring the babies through the various stages of screening, referral and confirmatory procedures. The HI*TRACK is a customised information management system for infant hearing screening programmes and it is widely used within and outside the USA. The researcher received hands-on training and technical support from NCHAM in USA on the use of the software. The researcher was assisted in this task by the data entry clerk who was well trained in the use of the HI*TRACK, particularly for generating summary reports for follow-up purposes. All HI*TRACK database were backed up in the researcher's personal laptop periodically. The researcher also maintained a separate diary for patients requiring follow-up along with their contact details.

3.12.3. Statistical Analysis

The data from the HI*TRACK was transferred to a spreadsheet (Microsoft Excel, 2003) for preliminary checking for any missing data. Thereafter, the data was exported to SPSS for Windows version 13.0 for all statistical analyses (SPSS Inc, Chicago, IL, USA). Cross-tabulation was used to provide a general overview of the results and a further check for missing data. Continuous variables such as age and birth weights of subjects were converted to categorical variables for ease of analysis. The first level of data analysis was with descriptive statistics of the explanatory and outcome variables. In the univariate analysis, the chi-squared test was used to describe the association between variables while odds ratio (OR) was stated

at 95% significance level. All tests of significance were two-tailed. Epi Info Version 3.4 (Centre for Disease Control, Atlanta, USA) was used to verify the results from the tests of significance for accuracy. Variables predictive of hearing impairment from putative risk factors were explored through case control analysis. This was accomplished in the hospital-based programme by matching children with hearing loss with children without hearing loss on the basis of age and sex. Because of the relatively small numbers of children with hearing loss, we chose the controls at a ratio of 1 to 10. This ratio was reduced to 1 to 5 for the community-based programme because of the larger number of children with hearing loss.

Multivariate analyses were performed with stepwise forward logistic regression to determine factors predictive of PCEHL among newborns in the hospital programme and among infants attending the community clinics for immunisation. This model was chosen because it allowed variables to be entered in a specified order one at a time based on the selected level of significance and it is particularly well-suited for reliably characterising the relationship between each predictor variable and the outcome variable [Katz, 2003]. Variables included in the logistic regression models were those with significance level of $p<0.1$. Although this threshold is higher than the default level ($p<0.05$) in the SPSS, it is considered more stringent than the 0.15 to 0.25 range proposed by Hosmer and Lemeshow [1989]. The results of the multivariate logistic regression analysis were presented in terms of adjusted odd ratios and the accompanying 95% confidence interval (CI).

3.12.4. Cost Analysis

The cost implications of any screening programme are vital for taking any health intervention to scale as a public health programme. Although the overriding objective of this project was to demonstrate the feasibility of infant hearing screening programmes in Nigeria, the cost of screening a baby under each programme was computed for a comparative analysis of the "cost-effectiveness" of the two screening models. Only fixed and variable direct costs were considered because of the limitations of determining all indirect

costs associated with the screening programmes in all the sites (Appendix 4.1). A detailed economic/cost-effectiveness analysis was also not feasible due to the lack of relevant data on disability-adjusted life years (DALYs) associated with PCEHL [Cook et al., 2006].

Moreover, screening performance may be affected by the sensitivity and specificity of the screening tests and protocol which in turn may affect costs in at least three ways [Davis, Bamford & Stevens, 2001]. Firstly, are the monetary and psychological costs of false negative outcomes which are difficult to determine. Secondly, there is the service cost of follow-up for children with false positive outcomes as well as the associated psychological costs. Thirdly, effective follow-up of children referred from the screening programme as well as the achievement of a high coverage often entails extra efforts that may also be difficult to quantify in monetary terms. Notwithstanding, the cost per screening a baby still provides a valuable measure of the "cost-effectiveness" of various screening options.

Chapter 4

Results

4 Results

4.1. Characteristics of the Study Participants

A total of 3,333 infants were screened throughout the duration of this project. Under the hospital-based programme, 1,330 babies were screened for a period of 40 weeks from Monday, May 16, 2005 to Saturday, February 18, 2006. Under the community-based programme, 2,003 infants were screened for a period of 40 weeks from Monday, July 4, 2005 to Thursday, April 6, 2006. The characteristics of the mothers and infants who participated in both programmes are presented in the following section.

4.1.1. Maternal Profile

4.1.1.1. Socio-Economic Status of Mothers

A summary of the socio-economic profile of mothers is presented in Table 4-1. Maternal age for all participants ranged from 15 to 48 years with a mean age of 29.3 years in the hospital-based programme and 28.0 years in the community-based programme. Majority (85.9%) of mothers were in the age group of 20 to 35 years. Very few mothers (3.2%) were teenagers (<20 years) while mothers above 35 years constituted 10.4% of the population. Overall, the ages of 17 mothers could not be ascertained. The age distribution of the mothers in each group is presented in Figures 4.1 and 4.2 below. The age patterns in both the hospital and the community populations were normally distributed.

Majority of mothers (97.1%) were married and consanguineous unions were rare. The Yoruba, the dominant ethnic group in the Southwest of Nigeria, constituted 83.7% of our total study population. Ibos from the Southeast and Hausas from the North were in the minority with 12.5% and 2.4% of the population respectively.

Table 4-1 Socio-Economic Status of Mothers

Profile	Hospital (%) n=1,330	Community (%) n=2,003	Total (%) n=3,333
Maternal Age (Years)			
<20	30 (2.2)	75 (3.7)	105 (3.2)
20 - 35	1,125 (84.6)	1,740 (86.9)	2,865 (85.9)
>35	171 (12.9)	175 (8.7)	346 (10.4)
Unknown	4 (0.3)	13 (0.7)	17 (0.5)
Range	15 – 46	15 – 48	
Mean ± SD	29.3 ± 5.1	28.0 ± 5.3	
Marital Status			
Single	48 (3.6)	45 (2.3)	93 (2.8)
Married	1,280 (96.2)	1,953 (97.5)	3,233 (96.9)
Widowed	2 (0.2)	0	2 (0.1)
Separated	0	0	0
Consanguinity	7 (0.5)	9 (0.5)	9 (0.5)
Ethnicity			
Yoruba	993 (74.7)	1,799 (89.8)	2,792 (83.7)
Ibo	274 (20.6)	142 (7.1)	416 (12.5)
Hausa	42 (3.2)	38 (1.9)	80 (2.4)
Other	21 (1.6)	24 (1.2)	45 (1.4)
Religion			
Christianity	776 (58.3)	556 (27.8)	1,332 (40.0)
Islam	553 (41.6)	1,442 (72.0)	1,995 (59.8)
Traditional	0	0	0
Other	1 (0.1)	0	1 (0)
Education			
None	37 (2.8)	32 (1.6)	69 (2.1)
Primary	97 (7.3)	269 (13.4)	366 (11.0)
Secondary	692 (52.0)	1,401 (70.0)	2,093 (62.8)
Tertiary	504 (37.9)	296 (14.8)	800 (24.0)
Education of Spouse			
None	32 (2.4)	13 (0.6)	45 (1.4)
Primary	37 (2.8)	78 (3.9)	115 (3.5)
Secondary	582 (43.8)	1,358 (67.8)	1,940 (58.3)
Tertiary	679 (51.0)	549 (27.4)	1,228 (36.8)
Occupation			
None	296 (22.3)	245 (12.2)	541 (16.2)
Small trade	423 (31.8)	1,196 (59.7)	1,619 (48.6)
Casual job	93 (7.0)	80 (4.0)	173 (5.2)
Full-time job	518 (39.0)	477 (23.8)	995 (29.8)
Occupation of Spouse			
None	52 (3.9)	45 (2.3)	97 (2.9)
Small trade	136 (10.2)	339 (16.9)	475 (14.3)
Casual job	69 (5.2)	69 (3.5)	138 (4.1)
Full-time job	1,073 (80.7)	1,501 (74.9)	2,574 (77.2)
Unknown	-	49 (2.4)	49 (1.5)
Accommodation			
Shared Toilets	1,268 (95.3)	1,934 (96.6)	3,202 (96.1)
Owned	134 (10.1)	143 (7.1)	277 (8.3)

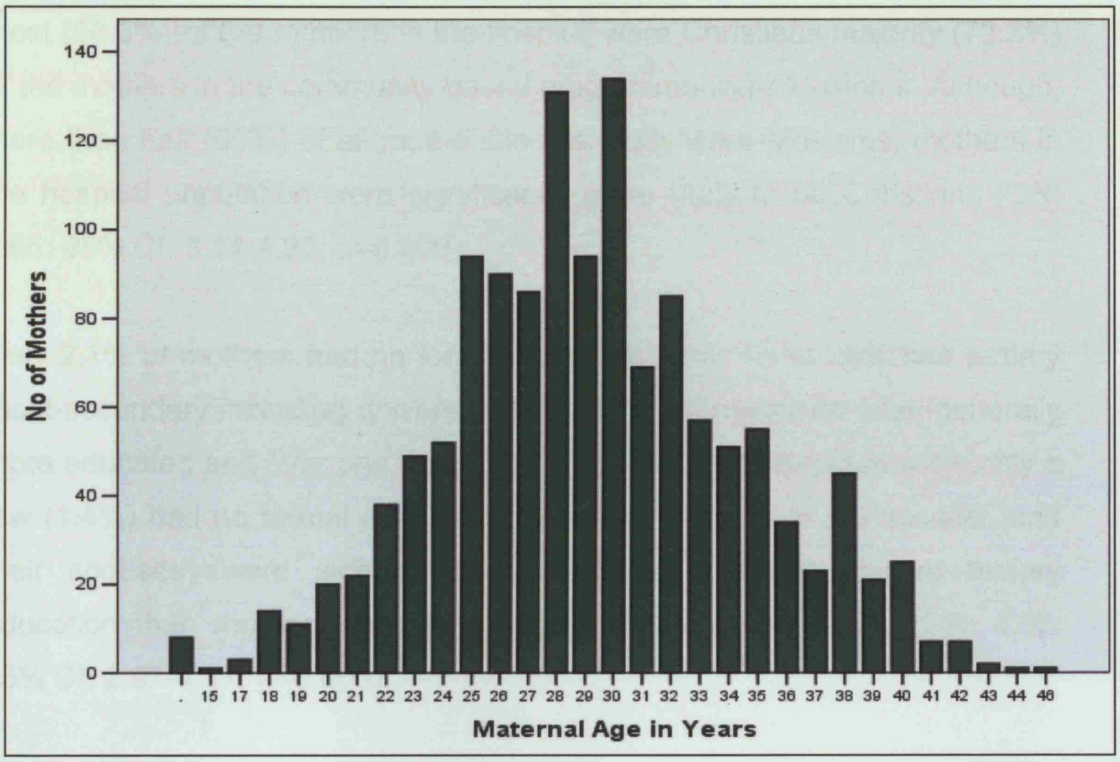


Figure 4.1. Age Distribution of Mothers in the Hospital

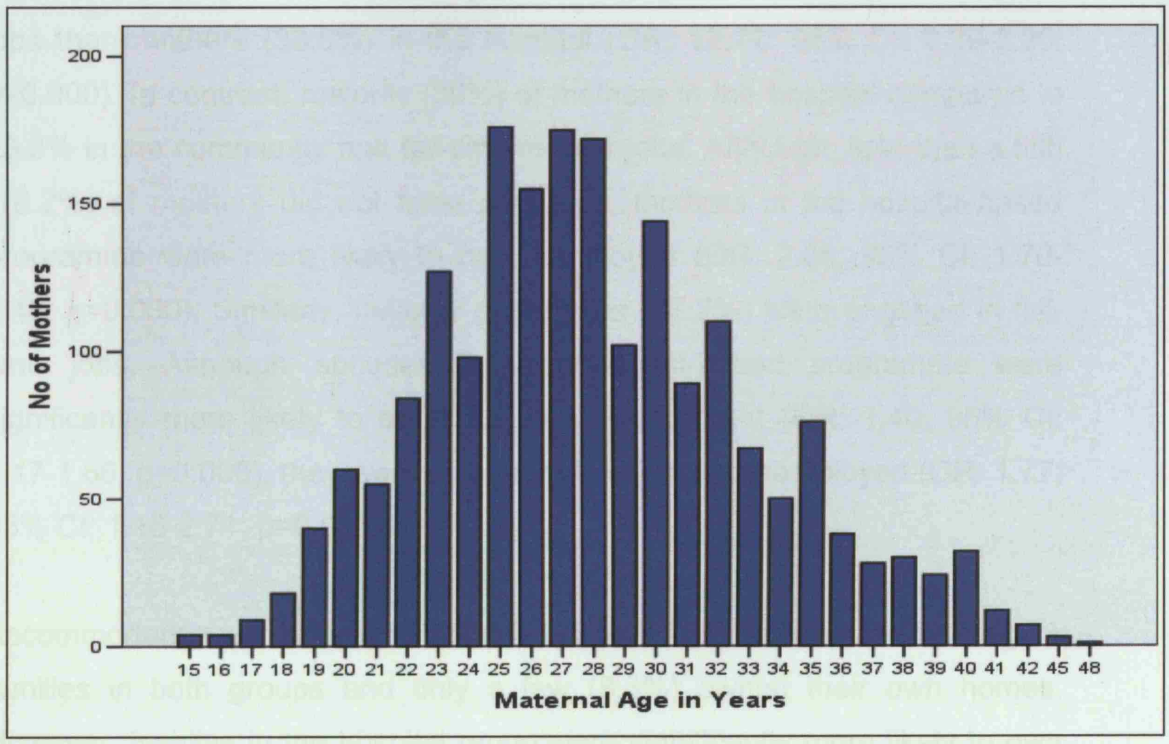


Figure 4.2. Age Distribution of Mothers in the Community

Religion was a significant part of the social life of women in this study. While most (58.3%) of the mothers in the hospital were Christians majority (72.2%) of the mothers in the community-based programme were Moslems. Although, more than half (60%) of all mothers in this study were Moslems, mothers in the hospital population were significantly more likely to be Christians (OR: 3.65; 95% CI: 3.14-4.23; p=0.000).

Only 2.1% of mothers had no formal education while up to 24% had tertiary (post-secondary including university) education. The spouses were generally more educated and over one third (36.8%) had tertiary education while only a few (1.4%) had no formal education. Mothers delivering in the hospital (and their spouses) were significantly more likely to have attained tertiary education than mothers (and their spouses) in the community (OR: 3.52; 95% CI: 2.97-4.17; p=0.000).

Most (63.7%) of the mothers employed in the community were engaged in small trade, casual or daily paid jobs. A high percentage of mothers (63.7%) in the community were more likely to be engaged in small trade or casual jobs than mothers (38.8%) in the hospital (OR: 12.77; 95% CI: 2.39-3.20; p=0.000). In contrast, majority (39%) of mothers in the hospital compared to 23.8% in the community had full-time regular jobs. Although, less than a fifth (16.2%) of mothers did not have paid jobs, mothers in the hospital-based programme were more likely to be unemployed (OR: 2.05; 95% CI: 1.70-2.48; p=0.000). Similarly, majority of spouses (77.2%) were engaged in full-time jobs. Although spouses in the hospital-based programme were significantly more likely to be in full-time employment (OR: 1.40; 95% CI: 1.17-1.66; p=0.000), they were also more likely to be unemployed (OR: 1.77; 95% CI: 1.16-2.71; p=0.005).

Accommodation with shared toilets (96.1%) was the most popular housing for families in both groups and only a few (8.3%) owned their own homes. However, families in the hospital group were significantly more likely to own their places of abode (OR: 1.46; 95% CI: 1.13-1.88; p=0.003).

4.1.1.2. Medical and Health-Seeking Profile

Table 4-2 summarises the key medical history and health-seeking behaviour of the mothers in this study. About half (50.4%) of the total population of mothers were multiparous. Although more mothers in the community (51.3%) were multiparous compared to the mothers in the hospital, the difference was not statistically significant (p=0.27).

Table 4-2. Maternal Medical and Health-Seeking Profile

Profile	Hospital (%) n=1,330	Community (%) n=2,003	Total (%) n=3,333
Parity			
Primiparous	674 (50.7)	970 (48.4)	1,644(49.3)
Multiparous	656 (49.3)	1,027 (51.3)	1,683 (50.5)
Missing	0	6 (0.3)	6 (0.2)
Pregnancy History			
Ante-natal care	867 (65.2)	1,988 (99.3)	2,855 (85.6)
Maternal malaria	241 (18.1)	395 (19.7)	636 (19.1)
Herbal drugs in pregnancy	263 (19.8)	1,364 (68.1)	1,627 (48.8)
Smoking	0	3 (0.1)	3 (0.1)
Alcohol intake	0	5 (0.2)	5 (0.2)
Co-existing Illness			
Maternal HIV	80 (6.0)	n/a	80 (2.4)
Pre-eclampsia	8 (0.6)	4 (0.2)	12 (0.4)
Eclampsia	45 (3.4)	3 (0.1)	48 (1.4)
Hypertension	2 (0.2)	4 (0.2)	6 (0.2)
Diabetes mellitus	3 (0.2)	0	3 (0.1)
Sickle cell disease	3 (0.2)	0	3 (0.1)
Place of Delivery			
Government Hospitals*	1,329 (100)	476 (23.8)	1,805 (54.2)
Private Hospital	-	422 (21.1)	422 (12.7)
Herbal Maternity Home	-	853 (42.5)	853 (25.6)
Family Home	-	119 (5.9)	119 (3.6)
Church	-	122 (6.1)	122 (3.7)
Born Before Arrival	1 (0)	6 (0.3)	7 (0.2)
Unknown	-	5 (0.2)	5 (0.2)
Mode of Delivery			
Spontaneous Vertex	696 (52.3)	1,904 (95.1)	2,600 (78.0)
Caesarean	575 (43.2)	92 (4.6)	667 (20.0)
Breech	32 (2.4)	0	32 (1.0)
Forceps/Vacuum	27 (2.1)	0	27 (0.8)
Unknown	0	7 (0.3)	7 (0.2)

*Health Centres included n/a: not available

A very notable pregnancy history in this study was the high uptake (85.6%) of antenatal care in both populations. Almost all mothers (99.3%) in the community had antenatal care compared to 65.2% in the hospital. In fact, mothers attending the immunisation clinics were significantly more likely to have received antenatal care than those who delivered in the hospital (OR: 70.78; 95% CI: 41.18-123.72; p=0.000). Another important finding was that nearly half of the mothers (48.8%) admitted to using herbal drugs in pregnancy. A significantly larger proportion (68.1%) of mothers in the community compared to 19.8% (263) in the hospital reported the use of herbal medications during pregnancy (OR: 8.66; 95% CI: 7.32-10.24; p=0.000). Further analysis revealed that about 62% of mothers who delivered in hospitals under the community-based programme had taken herbal drugs in pregnancy (data not shown). Maternal malaria was documented in a comparable proportion of mothers in the two groups (p=0.250). Alcohol and cigarette intake were generally uncommon in the study population. Although there was no reported life-style history of smoking or alcohol consumption among mothers in the hospital population, 3 women in the community admitted to smoking cigarettes and occasional/frequent intake of alcohol was documented for 5 others.

Maternal HIV was the most prevalent co-existing illness among mothers in the hospital group. A total of 80 mothers were HIV positive and 8 of the babies were admitted into the SCBU. Maternal history of HIV was excluded from the population of mothers in the community because their HIV status could not be verified. The next most predominant illness among the mothers in the hospital was eclampsia which was documented in 45 mothers.

The majority (55.2%) of mothers attending immunisation clinics did not deliver in hospitals (including all health facilities with maternity units). Detailed analysis of mothers' utilisation of maternity services in the community population revealed a startling preference for private non-hospital-based services (54.6%; n=1,094) over hospital/medical facilities

(45.1%; n= 904) with majority (42.5%; n= 853) using herbal maternity homes (Figure 4.3).

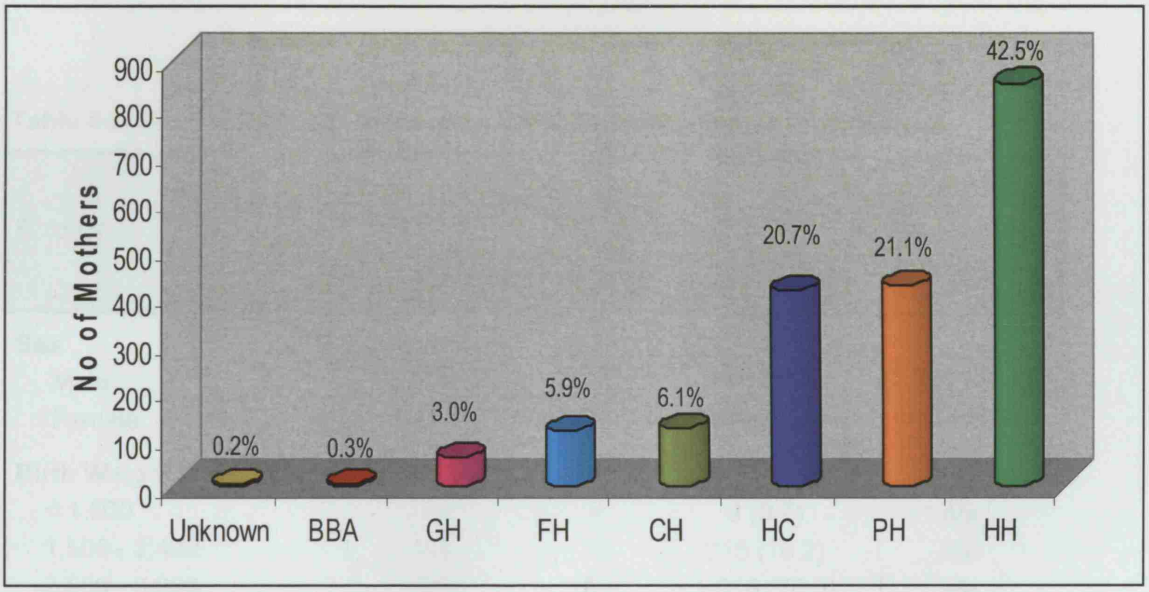


Figure 4.3. Uptake of Maternity Services by Mothers attending BCG Clinics

Key: BBA= Born before arrival; CH = Church; FH = Family home; HC = Health centre PH = Private Hospital; GH = Government Hospital; HH=Herbal Maternity Home

A smaller proportion (23.7%) of mothers opted for government owned/run health centres or hospitals compared with 69.6% who used private facilities outside their homes. Few mothers (0.3%) delivered before arrival in hospital and the places of birth for 5 abandoned babies were unknown. Virtually all the mothers who delivered outside hospital facilities received antenatal care in hospitals (data not shown), which further suggests a strong preference for the combined use of traditional and orthodox maternity services.

Majority of mothers in both groups delivered by spontaneous vertex. However, mothers in the community were significantly more likely to choose this mode of delivery than those in the hospital (OR: 17.52; 95% CI: 13.85-22.18; p=0.000). In contrast, 43.2% of the mothers in the hospital delivered their babies by caesarean section compared to 4.6% in the community and the difference was also statistically significant (OR: 15.82; 95% CI: 12.42-20.17; p=0.000). Delivery by assisted breech, forceps or vaccum extraction was not reported among mothers in the community.

4.1.2. Profile of Infants

A summary of the profile of infants screened during this project is presented in

Table 4-3. Profile of Infants in Hospital and Community-based Programmes

Profile	Hospital			Community n=2,003 (%)
	WBN n=1,150	SCBU n=180	Total n=1,330 (%)	
Sex				
Male	580	87	667 (50.2)	1,023 (51.1)
Female	570	93	663 (49.8)	980 (48.9)
Birth Weight (Grams)				
< 1,500	-	9	9 (0.7)	n/a
1,500 - 2,499	144	71	215 (16.2)	n/a
2,500 - 3,999	938	80	1,018 (76.5)	n/a
≥ 4000	51	14	65 (4.9)	n/a
Unknown	17	6	23 (1.7)	n/a
Gestational Age (Weeks)				
<34	23	32	55 (4.1)	15 (0.7)
≥34	1,112	146	1,258 (94.6)	1,981 (98.9)
Unknown	15	2	17 (1.3)	7 (0.3)
Multiple Gestation				
Singleton	1,066	151	1,217 (91.5)	1,997 (99.7)
Multiple	84	29	113 (8.5)	6 (0.3)
Place of Birth				
Herbal Home	0	0	0	853 (42.6)
Government Hospital	1,149	180	1,329 (99.9)	476 (23.8)
Private Hospital	0	0	0	422 (21.1)
Church	0	0	0	122 (6.1)
Family Home	0	0	0	119 (5.9)
Born Before Arrival	1	0	1 (0.1)	6 (0.3)
Unknown	0	0	0	5 (0.2)
Mode of Delivery				
Spontaneous Vertex	649	47	696 (52.3)	1,904 (95.1)
Caesarean	458	117	575 (43.2)	92 (4.6)
Breech	26	6	32 (2.4)	0
Forceps/Vacuum	17	10	27 (2.1)	0
Unknown	0	0	0	7 (0.3)

n/a: not available

Of the 1,330 babies screened in the hospital, 13.5% (180) were admitted into the SCBU while 2,003 infants were screened in the community programme. The male populations (50.2% vs. 51.1%) in both groups were comparable and slightly more than the female populations (49.8% vs. 48.9%). Birth weights in the hospital varied from 1.0 to 5.9 kg and were slightly skewed beyond the mean of 3.06 kg (Figure 4.4).

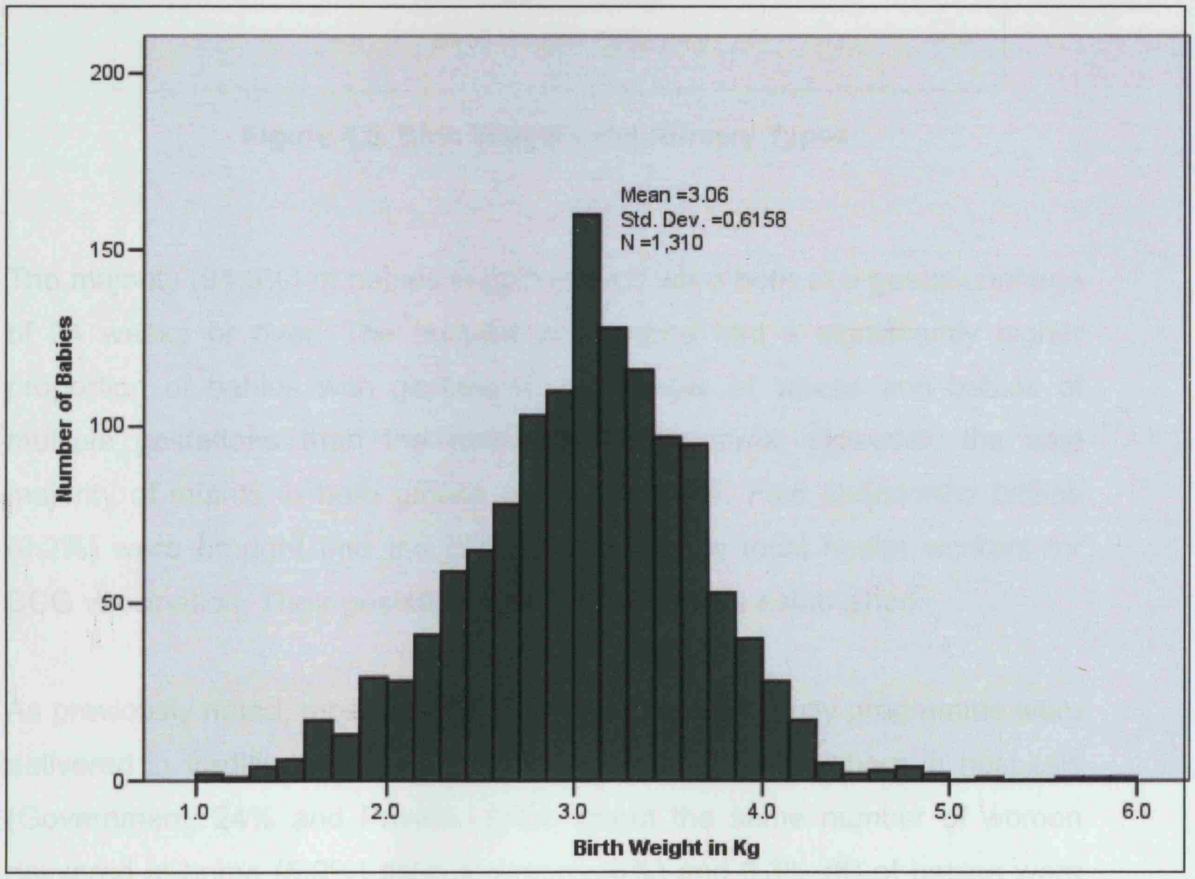


Figure 4.4. Distribution of Birth Weights of Babies in the Hospital

All babies with very low-birth weights (<1.5 kg) as well as a significant number (33 - 34%) of babies weighing <2.5 kg and >4.0 kg required SCBU admission compared to babies with normal birth weights (Figure 4.5). Comparable data on birth weights for infants in the community was not readily available.

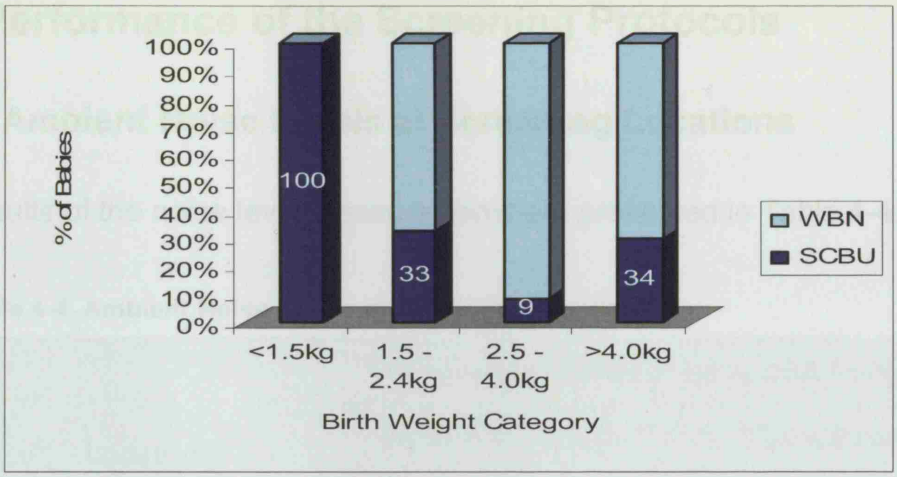


Figure 4.5. Birth Weights and Nursery Types

The majority (94.6%) of babies in both groups were born at a gestational age of 34 weeks or over. The hospital programme had a significantly higher proportion of babies with gestational age below 34 weeks and babies of multiple gestations than the community programme. However, the vast majority of infants in both groups were singletons. Five abandoned babies (0.2%) were brought into the community clinic by local health workers for BCG vaccination. Their gestational ages could not be established.

As previously noted, most babies (42.5%) in the community programme were delivered in traditional maternity homes. While 45% were born in hospitals (Government, 24% and Private, 21%), about the same number of women delivered at home (5.9%) as in churches (6%) and 0.3% (6) of babies were born before arrival in hospital. Majority (65%) of the babies in SCBU were delivered by caesarean section compared to about 40% in the WBN (p=0.000). Overall, 43.2% of the babies in the hospital were delivered by caesarean section compared to 4.6% in the community (p=0.000). The vast majority (95.1%) of infants in the community were delivered by spontaneous vertex.

4.2. Performance of the Screening Protocols

4.2.1. Ambient Noise Levels at Screening Locations

The results of the noise level measurements are presented in Table 4-4.

Table 4-4. Ambient Noise Levels at Screening Locations

Location		Average Noise Levels in dBA (range)	
		Open Ward/Clinic	Quiet Room
Hospital	WBN	66.9 (61.0 - 76.0)	60.3 (52.5 - 74.0)
	SCBU	76.9 (62.3 - 90.5)	65.4 (59.8 - 67.5)
Community	BCG Clinic 1	65.4 (60.5 - 78.4)	63.1 (59.0 - 69.5)
	BCG Clinic 2*	64.1 (59.3 - 83.1)	63.4 (55.6 - 74.5)
	BCG Clinic 3	66.9 (61.0 - 76.0)	65.3 (58.8 - 69.8)
	BCG Clinic 4	75.6 (61.4 - 89.4)	67.9 (60.3 - 82.5)

*Location for all AABR tests in the community-based programme

4.2.1.1. Hospital-based Programme

The average ambient noise level in the SCBU was 76.9 dBA (62.3 to 90.5 dBA) and 66.9 dBA (61.0 to 76.0 dBA), in the WBN. The quiet rooms in the lying in ward and the SCBU were the least noisy spaces available for screening. Typical noise levels in the quiet rooms were 65.4 (59.8 to 67.5) dBA in the SCBU and 60.3 (52.5 to 74.0) dBA in the WBN.

4.2.1.2. Community-based Programme

The ambient noise levels ranged from 64.1 to 75.6 dBA because of the location of the clinics in busy areas. The ambient noise levels in the quietest rooms available for screening in each of the 4 centres ranged from 63.1 to 67.9 dBA which were generally comparable to the quiet location in the SCBU in the hospital programme.

4.2.2. Screening Coverage

4.2.2.1. Hospital-based Coverage

The total number of births during the period of screening was 1,633. There were 259 (15.9%) still births and 1,374 live births (84.1%) which translated to a still-birth rate of 189 per thousand live-births in this population (Fig.4.6).

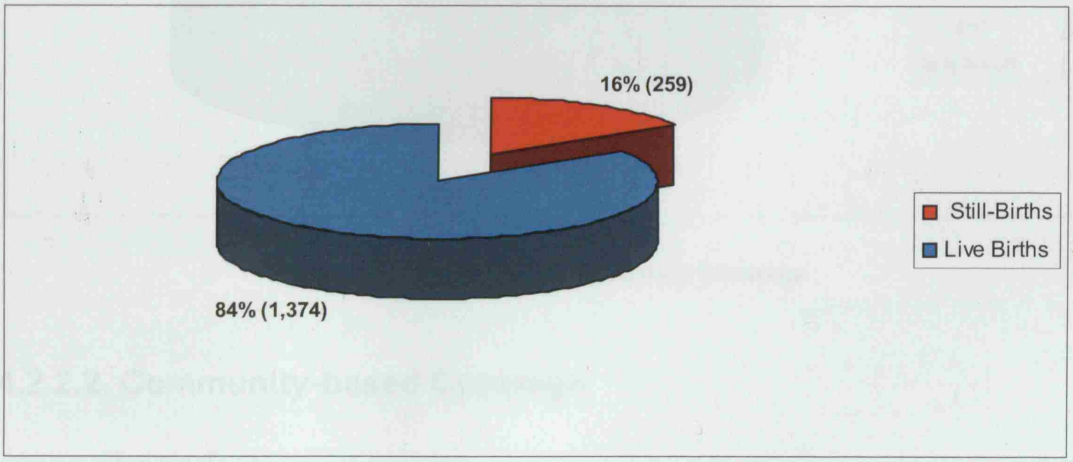


Figure 4.6. Hospital Birth Profile

Of the 1,374 live births, 2.0% (27 babies) died in the SCBU during the neonatal period, giving an in-patient neonatal mortality rate of 20 per 1,000 live births and a perinatal mortality rate of 208 per 1,000 live births. From the 1,347 survivors, 1.3% (17) babies missed the hearing screening leaving a balance of 1,330 babies. Hence, the screening coverage was 98.7%. Of the 17 babies that were not available for screening, 12 were transferred from the

SCBU to a nearby children's hospital because they needed urgent specialised clinical attention.

From the 1,330 babies screened under the hospital-based programme, 98% (1,304) were screened as in-patients (before hospital discharge), while the rest (26) who missed the in-patient screening were screened as out-patients after hospital discharge (Figure 4.7). Of the 1,304 babies screened before hospital discharge, 86.5% (1,128) were screened in WBN and 13.5% (176) in SCBU. Four of the 26 babies who missed the in-patient 1st stage screening were SCBU graduates.

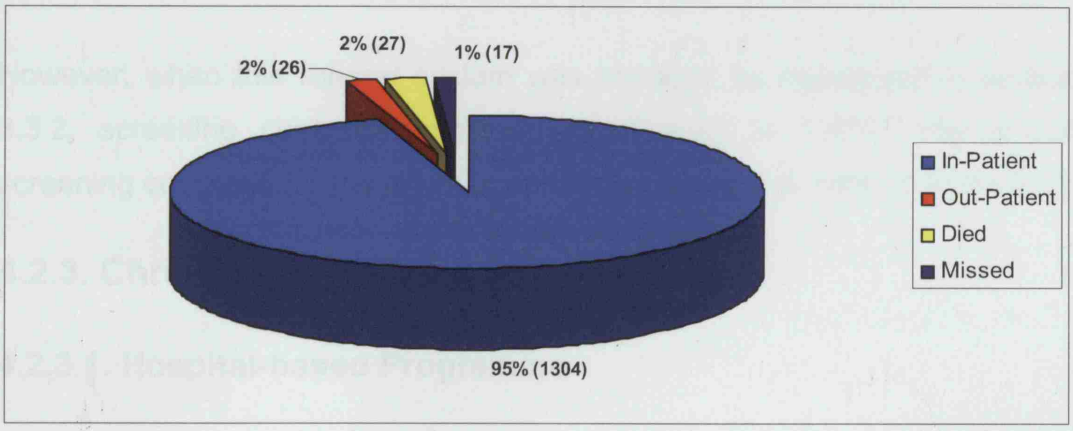


Figure 4.7. Hospital-based Screening Coverage

4.2.2.2. Community-based Coverage

A total of 2,991 babies attended the four primary health care centres for BCG immunisation during the study period. About 76% of the babies (2,277), who were less than 3 months old, were eligible for hearing screening as older babies (714) were excluded. Of the 2,277 eligible babies, 12% (274) missed the hearing screening and this occurred only in the first two months as a result of the initial referral system to a single clinic for hearing screening (Figure 4.8).

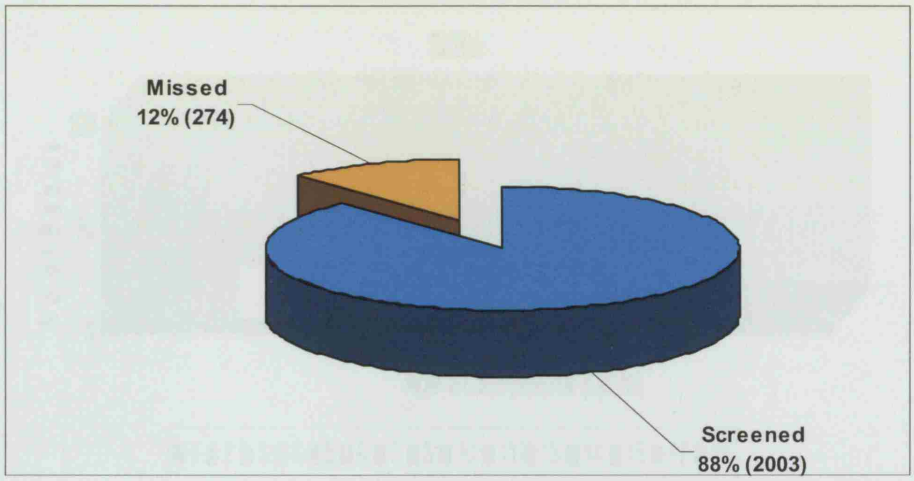


Figure 4.8. Community-based Screening Coverage

However, when this referral system was changed as highlighted in section 3.3.2, screening coverage improved significantly to 100%. The overall screening coverage for the entire period of screening was 88% (2,003/2,277).

4.2.3. Chronological Age at Screening

4.2.3.1. Hospital-based Programme

SCBU babies were mostly screened after the second day compared with the well babies (89.2%) who were screened by the second day of life (Fig 4.9). Babies screened during the out-patient clinic appointments were predominantly much older than 7 days and generally more than 42 days old. The mean ages of screening in the WBN, SCBU and OPD were 1.3, 3.5, and 54.3 days respectively. Overall, the mean age of screening in the hospital was 2.6 days (SD: 7.2; range: 1 – 104 days).

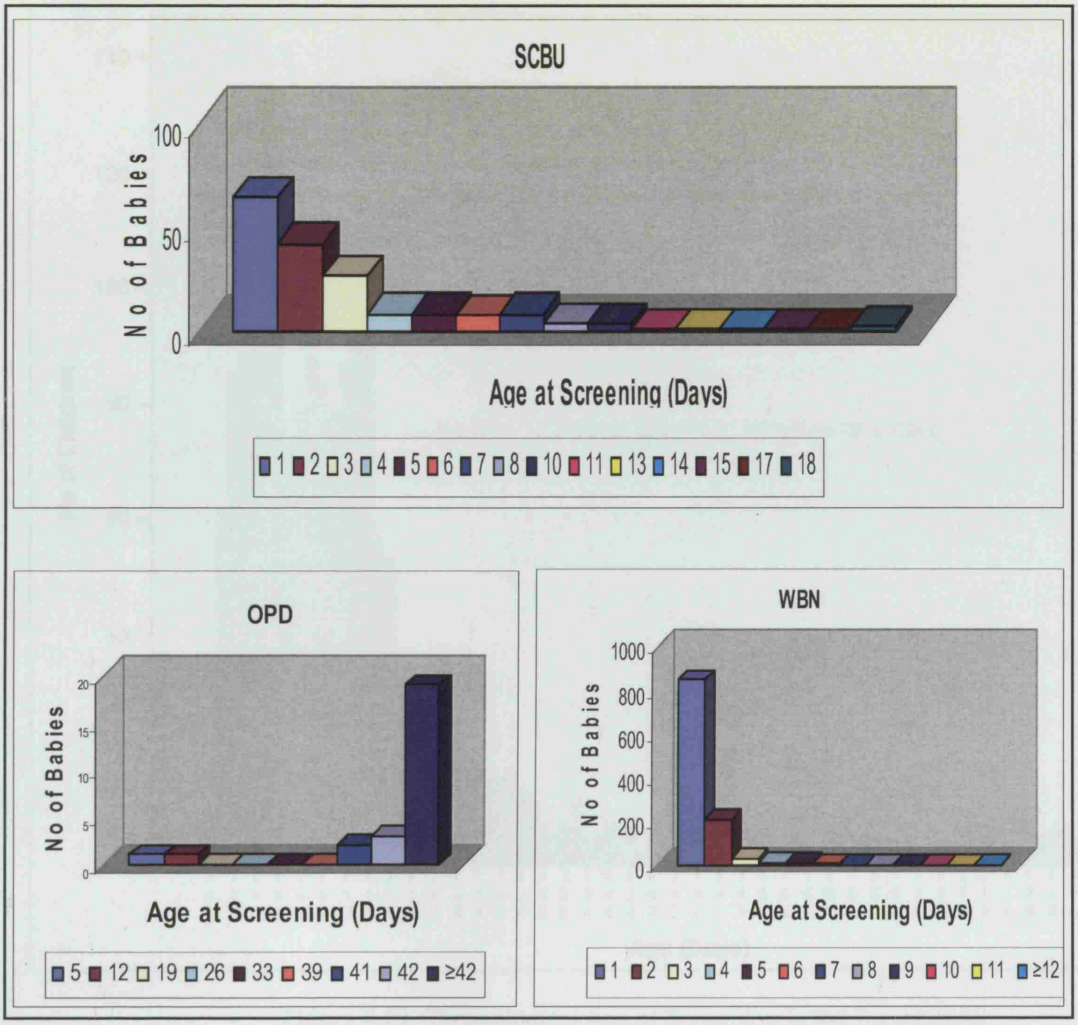


Figure 4.9. Chronological Age of Screening Babies in the Hospital

4.2.3.1. Community-based Programme

The age at screening for babies attending BCG clinic varied from 1 to 90 days with a mean of 17.7 days and standard deviation of 19.1 days (Figure 4.10.). The 90th percentile was 46.6 days. This distribution shows a fairly good uptake (over 75%) for BCG immunisation in the first month of life.

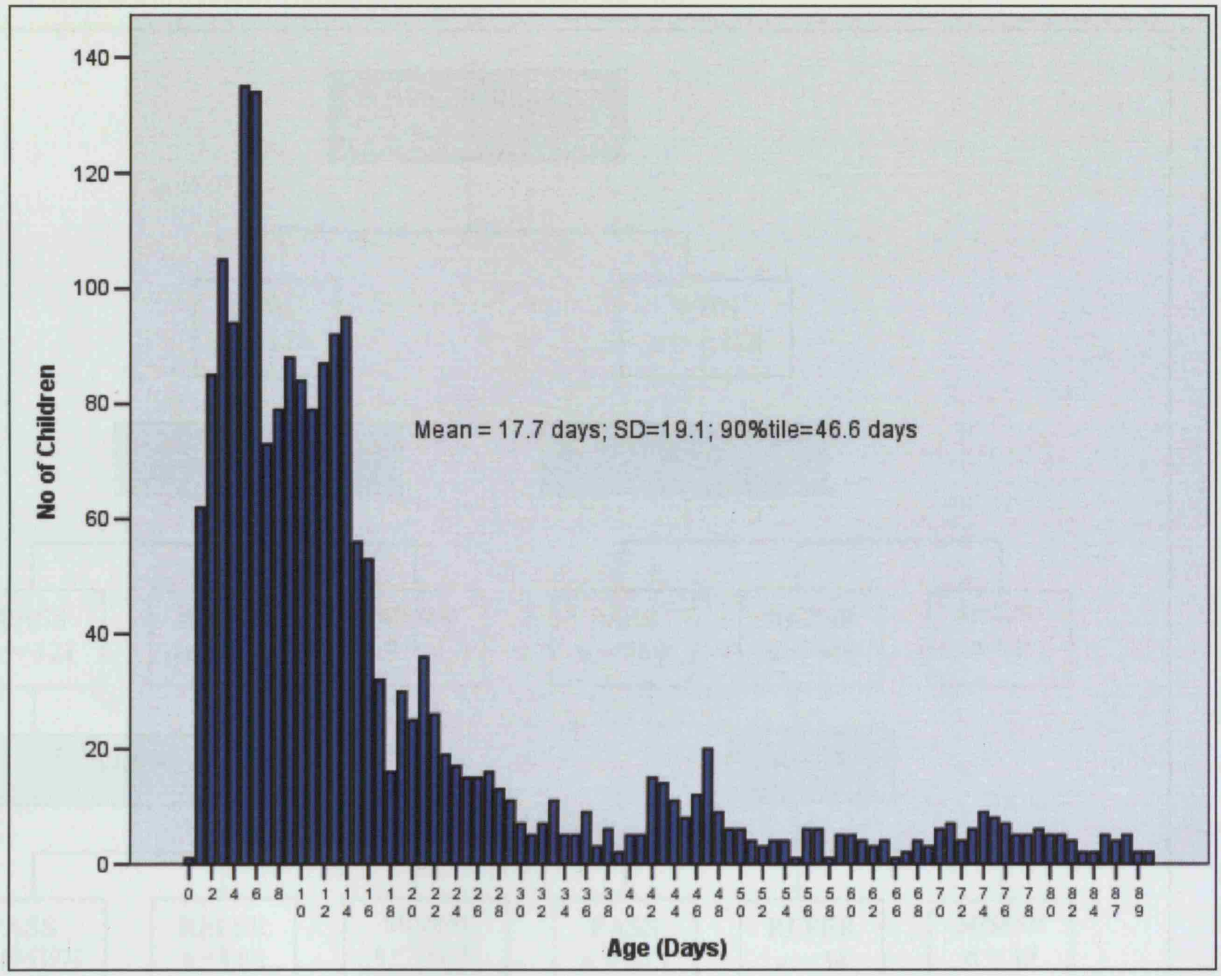


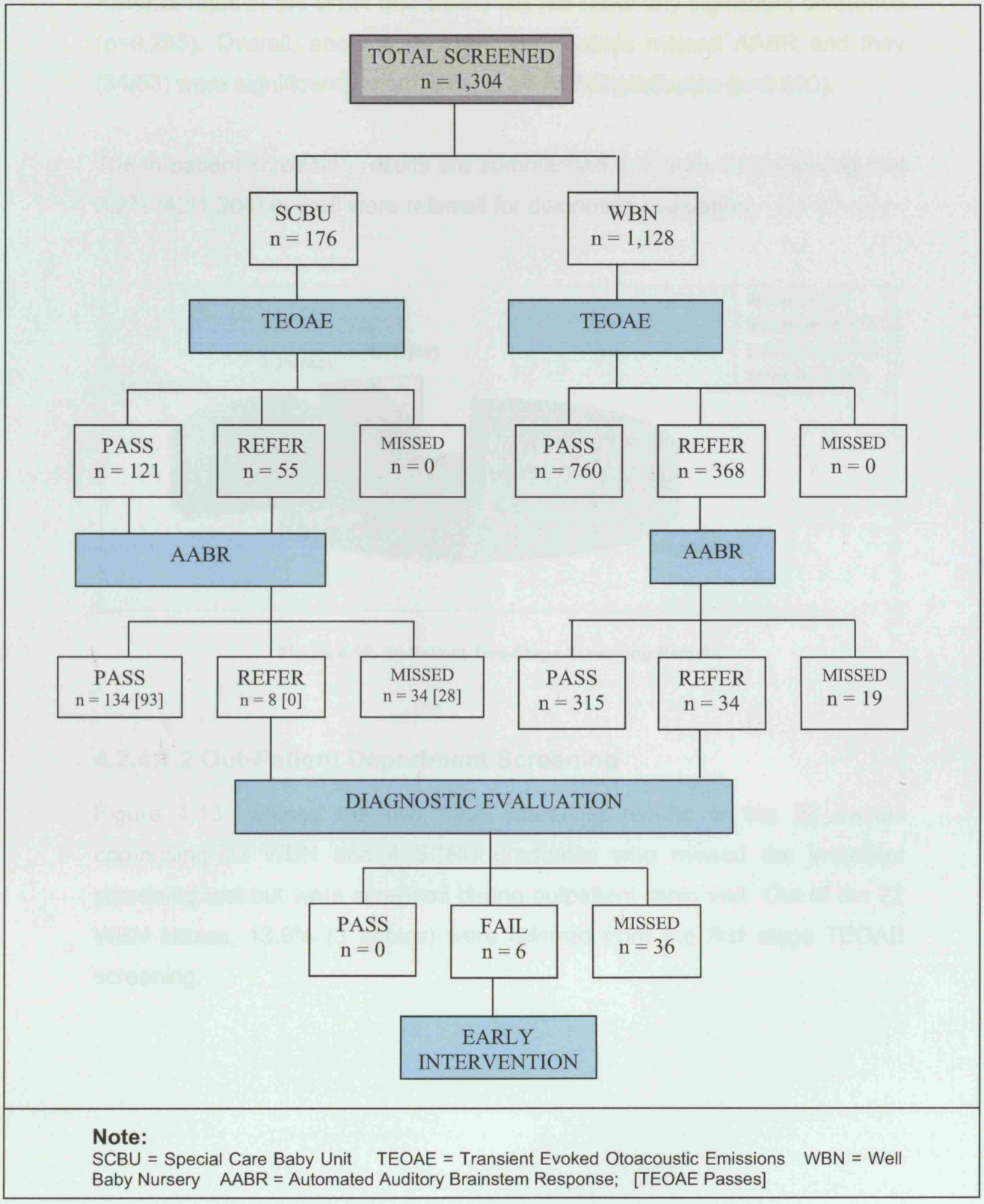
Figure 4.10. Chronological Age at Screening in the Community

4.2.4. Referral Rates

4.2.4.1. Hospital-based Programme

4.2.4.1.1. In-Patient Screening Results

The summary of neonates passing through the hospital screening programme is outlined in Figure 4.11. Out of the 1,128 babies screened in the WBN, 32.6% (368) babies were referred at the first stage TEOAE screening, 95% (349/368) had AABR and 9.7% (34/349) of them were referred. Overall, 3% (34/1,128) of the total population of well babies screened as in-patients were referred. In contrast, 4.5% (8/176) of SCBU babies were referred for diagnostic evaluation after AABR referral.



Note:
SCBU = Special Care Baby Unit TEOAE = Transient Evoked Otoacoustic Emissions WBN = Well Baby Nursery AABR = Automated Auditory Brainstem Response; [TEOAE Passes]

Figure 4.11. Summary of the In-patient Hospital-based Screening

Referral rates in the WBN and SCBU did not show any significant difference (p=0.285). Overall, about 4.1% (53/1,304) babies missed AABR and they (34/53) were significantly more likely to be SCBU graduates (p=0.000).

The in-patient screening results are summarised in Figure 4.12 showing that 3.2% (42/1,304) overall were referred for diagnostic evaluation.

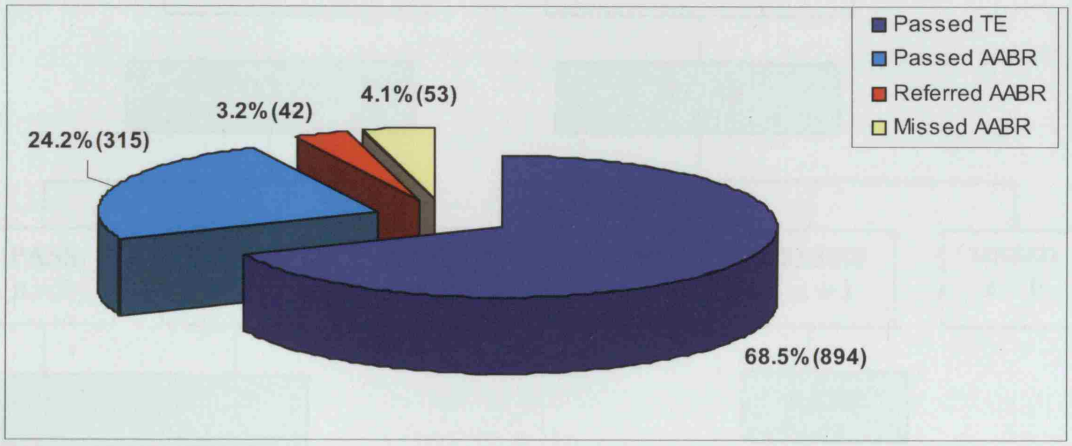
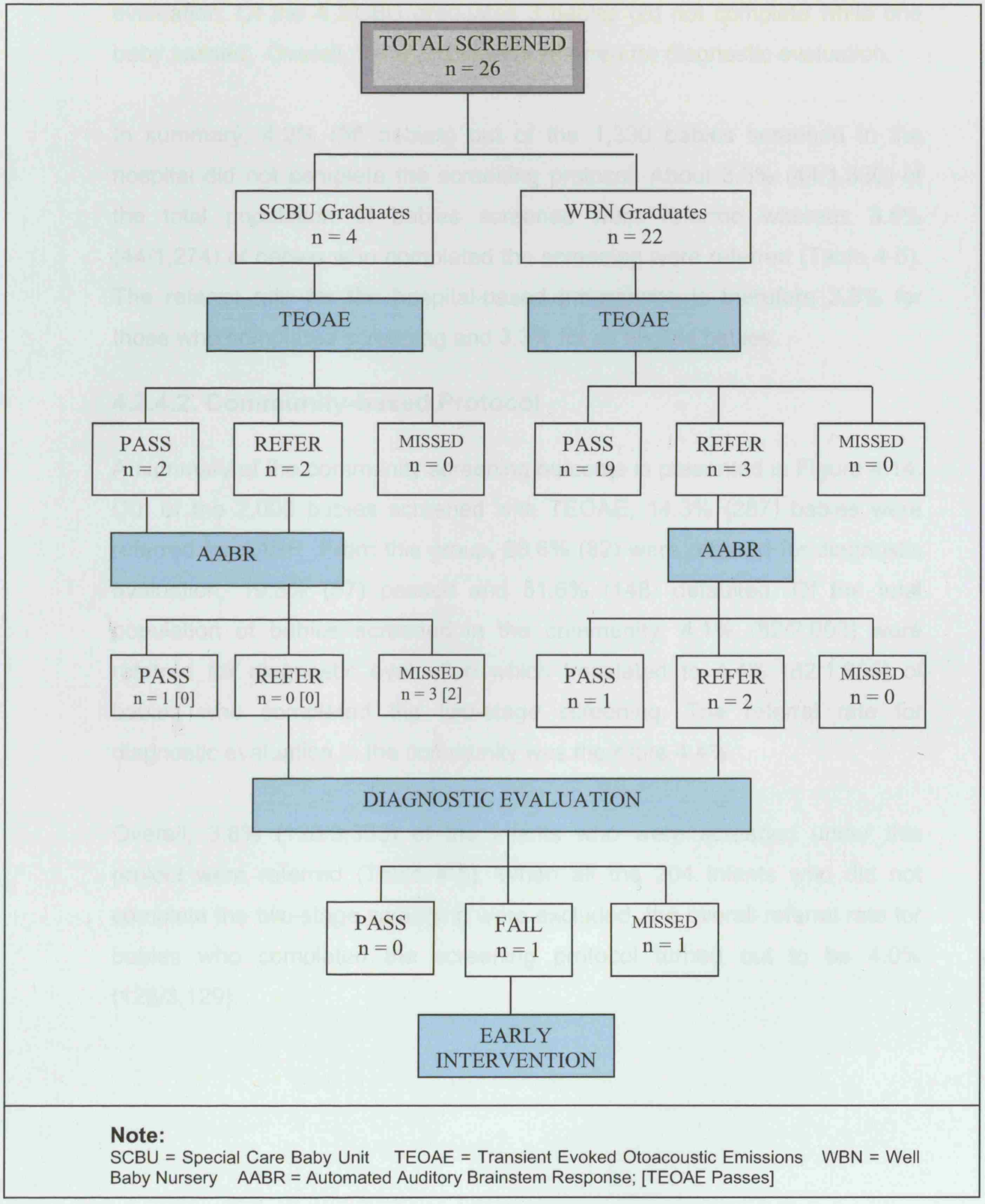


Figure 4.12. In-Patient Two-Stage Screening Results

4.2.4.1.2 Out-Patient Department Screening

Figure 4.13 outlines the two-stage screening results of the 26 babies comprising 22 WBN and 4 SCBU graduates who missed the in-patient screening test but were screened during outpatient clinic visit. Out of the 22 WBN babies, 13.6% (3 babies) were referred from the first stage TEOAE screening.



Note:
SCBU = Special Care Baby Unit TEOAE = Transient Evoked Otoacoustic Emissions WBN = Well Baby Nursery AABR = Automated Auditory Brainstem Response; [TEOAE Passes]

Figure 4.13. Summary of the Outpatient Hospital-based Screening for Babies who missed Inpatient Screening

Two thirds of babies (2/3) who had AABR were referred for diagnostic evaluation. Of the 4 SCBU graduates 3 babies did not complete while one baby passed. Overall, 7.7% (2/26) were referred for diagnostic evaluation.

In summary, 4.2% (56 babies) out of the 1,330 babies screened in the hospital did not complete the screening protocol. About 3.3% (44/1,330) of the total population of babies screened were referred whereas 3.5% (44/1,274) of babies who completed the screening were referred (Table 4-5). The referral rate for the hospital-based programme is therefore 3.5% for those who completed screening and 3.3% for all eligible babies.

4.2.4.2. Community-based Protocol

A summary of the community screening outcome is presented in Figure 4.14. Out of the 2,003 babies screened with TEOAE, 14.3% (287) babies were referred for AABR. From this group, 28.6% (82) were referred for diagnostic evaluation, 19.8% (57) passed and 51.6% (148) defaulted. Of the total population of babies screened in the community, 4.1% (82/2,003) were referred for diagnostic evaluation which translated to 4.4% (82/1,855) of babies who completed the two-stage screening. The referral rate for diagnostic evaluation in the community was therefore 4.4%.

Overall, 3.8% (126/3,333) of the infants who were screened under this project were referred (Table 4-5). When all the 204 infants who did not complete the two-stage screening were excluded, the overall referral rate for babies who completed the screening protocol turned out to be 4.0% (126/3,129).

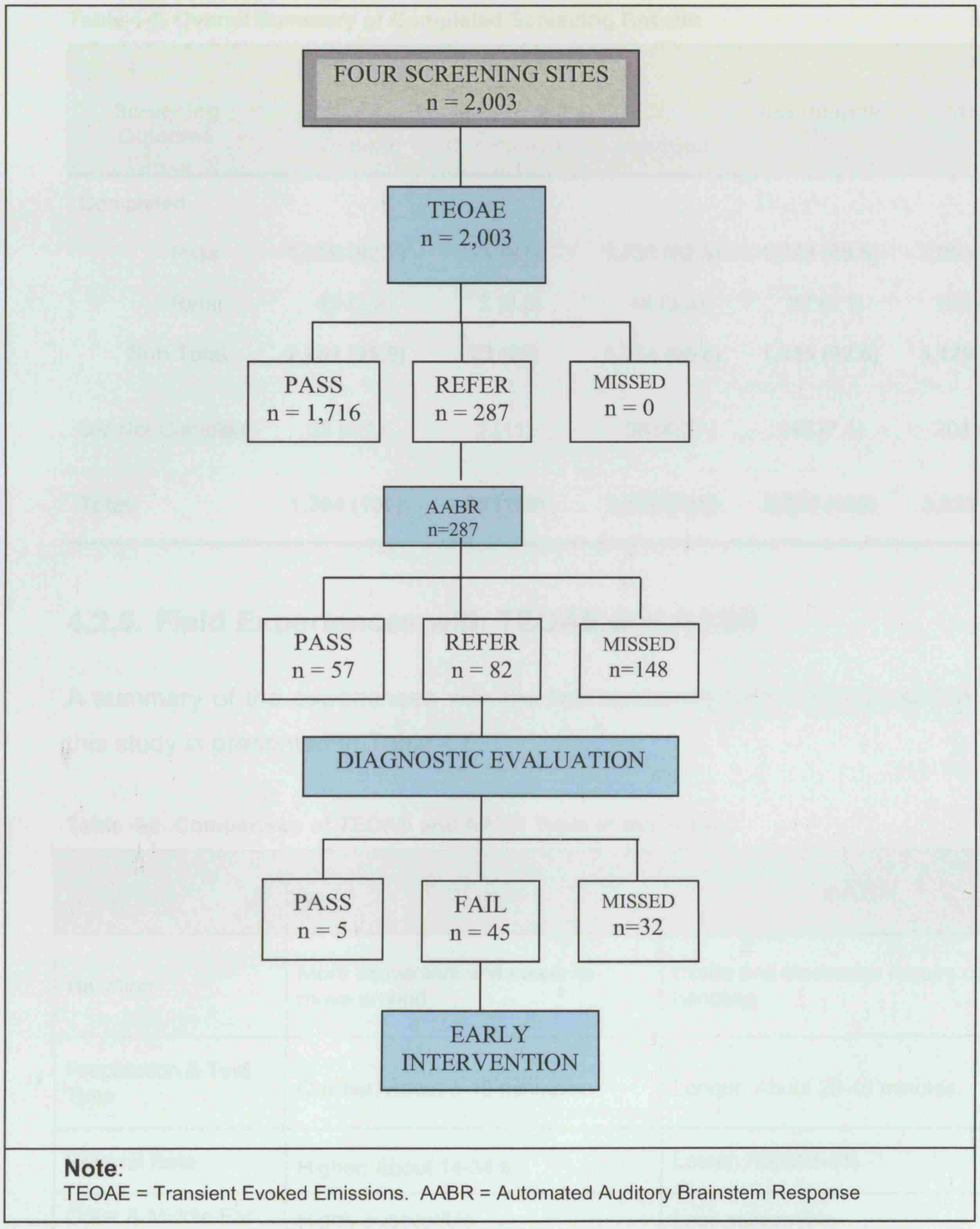


Figure 4.14. Summary of Community-based Screening

Table 4-5. Overall Summary of Completed Screening Results

Screening Outcome	Hospital-Based (%)			Community (%)	Total (%)
	In-Patient	Out-Patient	Sub-total		
Completed					
Pass	1,209 (92.7)	21 (81)	1,230 (92.5)	1,773 (88.5)	3,003 (90.1)
Refer	42 (3.2)	2 (8.0)	44 (3.3)	82 (4.1)	126 (3.8)
Sub Total	**1,251 (95.9)**	**23 (89)**	**1,274 (95.8)**	**1,855 (92.6)**	**3,129 (93.9)**
Did Not Complete	53 (4.1)	3 (11)	56 (4.2)	148 (7.4)	204 (6.1)
Total	**1,304 (100)**	**26 (100)**	**1,330 (100)**	**2,003 (100)**	**3,333 (100)**

4.2.5. Field Experiences with TEOAE and AABR

A summary of the experiences with the two screening technologies used in this study is presented in Table 4-6.

Table 4-6. Comparison of TEOAE and AABR Tests in this Study

Criterion	TEOAE	AABR
Handling	More convenient and easier to move around	Cords and electrodes require careful handling
Preparation & Test Time	Quicker: About 5-10 minutes	Longer: About 20-45 minutes.
Referral Rate	Higher: About 14-34%	Lower: About 3-4%
Outer & Middle Ear Obstruction	Highly susceptible	Less susceptible
Environmental Noise	Quiet baby and quiet test environment essential	Not a limiting factor.
False Negatives	Auditory Neuropathy will be missed	Mild PCEHL and high frequency hearing loss will be missed.

The preparation time was longer for AABR because of the need to scrub baby for electrode placement. Test time for TEOAE was quick, especially for a quiet baby. Total time for baby preparation, testing, and documentation was usually 5-10 minutes for TEOAE compared with 20-45 minutes for AABR. Referral rate for TEOAE of 14 to 34% was higher than 3 to 4.5% for AABR. While the cost of disposables for TEOAE at $0.10 per child was far less than the $12.00/child for AABR. However, TEOAE is more susceptible to outer ear vernix or debris and middle ear effusion and environmental noise than AABR. TEOAE also recorded false negative results from babies with auditory neuropathy while AABR could have missed minimal and high frequency hearing losses.

4.2.6. Return Rates for Diagnostic Evaluation

Only 15.9% (7/44) of the babies who were referred for diagnostic evaluation in the hospital returned compared with 61.0% (50/82) of the babies referred in the community. The return rate for diagnostic evaluation for the hospital-based study of 15.9% differed significantly from the 61.0% reported from the community-based programme (p=0.000).

4.2.7. Age at Confirmation

4.2.7.1. Hospital-based Programme

The age of diagnosis for the 7 babies with hearing loss in the hospital group varied widely from 46 to 360 days with a mean of 233 days (SD: 118.2 days). Only two children assessed during outpatient post-natal clinic, were confirmed with hearing loss within 3 months of life. One was a graduate of SCBU and the other was from the WBN. The others were confirmed well after 6 months as a result of an intensive/active follow-up of the parents. Two infants from the SCBU had been sent to the village to live with their grandparents because of their special needs, one of which was due to maternal death.

4.2.7.2. Community-based Programme

The age of diagnosis in the community ranged from 20 to 129 days (Figure 4.15) and the mean age of diagnosis was 51 days (SD: 29.4 days). This data is based on the total number of infants (n=56) confirmed with hearing loss in the community as detailed in section 4.2.8.2 below.

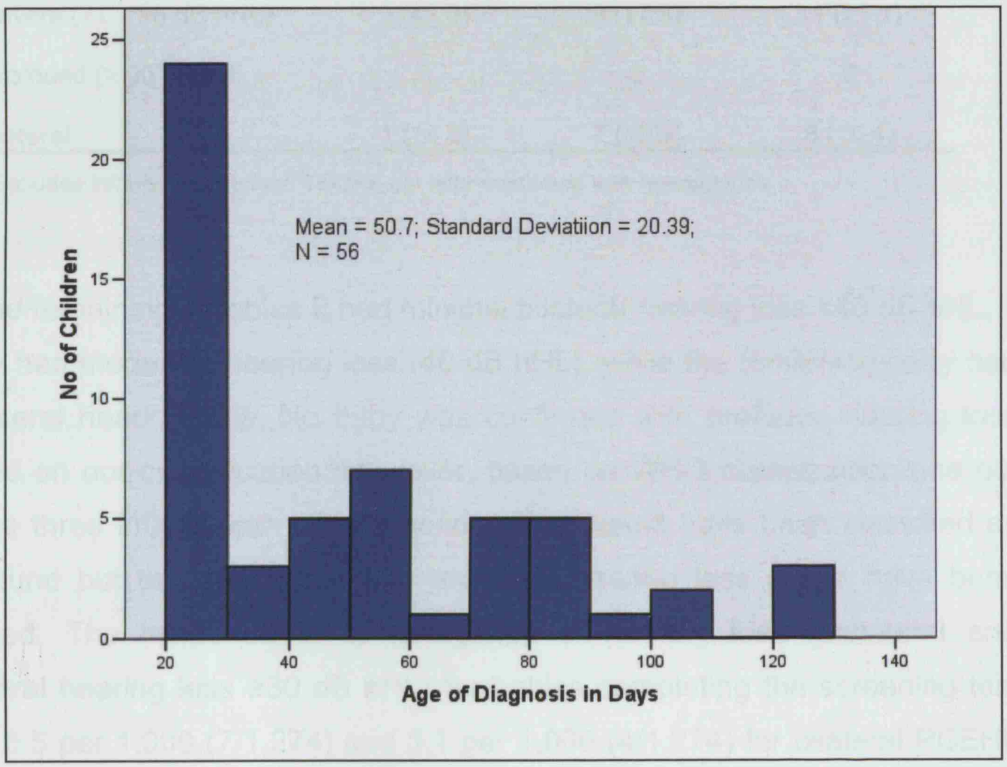


Figure 4.15. Age of Diagnosis of Hearing Loss in the Community

4.2.8. Summary of Diagnostic Results

The summary of the key outcomes after diagnostic evaluation is presented in Table 4-7.

4.2.8.1. Hospital-based Programme

All the 7 babies who returned for diagnostic evaluation out of the 44 who were referred after AABR were confirmed with hearing loss. Of these 7 babies, 3 were confirmed with severe hearing loss ≥ 70 dB nHL and all 3 were SCBU graduates.

Table 4-7. Summary of Diagnostic Results based on Screening Protocols

Type of Hearing Loss	Hospital n=7 (%)	Community* n=45 (%)	Total n=52 (%)
Bilateral			
Mild (30 - 40 dB nHL)	2 (28.6)	11 (24.4)	13 (25.0)
Moderate (40 – 70 dB nHL)	1 (14.3)	19 (42.2)	20 (38.5)
Severe (71 – 90 dB nHL)	3 (42.8)	8 (17.8)	11 (21.1)
Profound (> 90 dB nHL)	0	0	0
Unilateral	1 (14.3)	7 (15.6)	8 (15.4)

*Excludes infants who passed TEOAE but later confirmed with hearing loss

Of the remaining 4 babies 2 had minimal bilateral hearing loss <40 dB nHL, 1 baby had moderate hearing loss (40 dB nHL), while the remaining baby had unilateral hearing loss. No baby was confirmed with profound hearing loss based on our classification. However, based on WHO classification one out of the three infants with severe hearing loss would have been classified as profound but the one infant with unilateral hearing loss would have been missed. The incidence of all categories of hearing loss (unilateral and bilateral hearing loss ≥30 dB nHL) for babies completing the screening test was 5.5 per 1,000 (7/1,274) and 3.1 per 1,000 (4/1,274) for bilateral PCEHL ≥40 dB nHL. Similarly, the incidence among SCBU graduates was 21.0 per 1,000 (3/143) after adjusting for those who did not complete the two-stage screening. However, based on the total number of babies enrolled for the programme (n=1,330), the yield for PCEHL was 5.3 per 1,000 for all categories of hearing loss or 3.0 per 1,000 for PCEHL ≥40 dB nHL. Similarly, the yield for PCEHL in SCBU babies (n=180) was 16.7 per 1,000.

None of the 123 babies who passed TEOAE (representing 13.6% of all TEOAE passes) scheduled for re-testing with AABR was confirmed with hearing loss barring the 30 babies in this group who were missed in the in-patient and out-patient screening.

4.2.8.2. Community-based Programme

Of the 50 babies who returned for diagnostic assessment 90% (45) were confirmed with various degrees of PCEHL as shown in Table 4-7. Hearing loss was bilateral and severe in 17.8% (8/45), moderate in 42.2% (19/45), mild in 24.4% (11/45) but unilateral in 15.6% (7/45). Thus, the prevalence of all categories of hearing loss for babies completing the screening test was 24.3 per 1,000 (45/1,855) while the prevalence of hearing loss that is moderate to severe was 14.6 per 1,000 (27/1,855).

From the 1,716 babies who passed TEOAE screen in the community, 10% (172 babies) were scheduled for AABR test compared to 13.6% (123/902) in the hospital population drawn exclusively from SCBU. A total of 75 (44%) babies out of the 172 babies returned for AABR and 14 babies (18.7%) were referred for diagnostic evaluation out of which 11 (78.6%) were confirmed with hearing loss. These 11 babies who initially had passed TEOAE therefore constitute false negatives for the community-based programme. The degree of hearing loss was severe in 2, moderate in 6, mild in 1 and unilateral in 2 babies. No baby was confirmed with profound hearing loss based on our classification. In total, hearing loss was severe in 10, moderate in 25, mild in 12 and unilateral in 9 infants (Figure 4.16). However, based on WHO classification three out of the ten infants with severe hearing loss would have been classified as profound while all the nine infants with unilateral hearing loss would have been missed. After adjusting for those who did not complete the screening, the overall prevalence in the community was 30.2 per 1,000 (56/1,855). Alternatively, based on the total enrolment, the "yield" of PCEHL in the community was 28.0 per 1,000 (56/2,003). The prevalence of moderate-to-severe hearing loss was 18.7 per 1,000 (35/1,855). More males (36) were confirmed with hearing loss than females (20) and the difference was statistically significant (p=0.045).

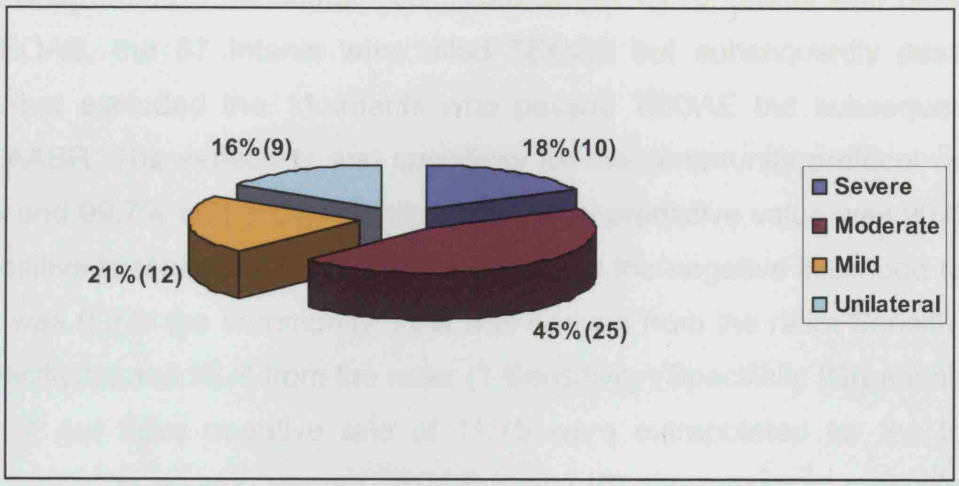


Figure 4.16. Summary of Diagnostic Evaluation for Community-based Infants

4.2.9. Efficiency of Screening Protocols

Summary of outcomes after diagnostic evaluation is presented in Table 4-8.

Table 4-8. Summary of Diagnostic Outcomes

Measure	Hospital (%) n=1,330	Community (%) n=2,003
True Positives	7 (0.5)	45 (2.2)
False Positives	0	5 (0.2)
True Negatives	n/a	1,762 (88.0)
False Negatives	n/a	11 (0.5)
All Missed Tests	93 (7.0)	180 (9.0)

n/a: not available

The very low return rate for diagnostic evaluation of 15.9% (7/44) in the hospital population compared with 61% (50/82) in the community would make the estimates of sensitivity, specificity and positive predictive value derivable from the outcomes in Table 4-8 unrealistic and were therefore not explored further.

A total of 1,762 infants were classified as true negatives in the community-based programme. This number consisted of the 1,716 infants who passed the TEOAE, the 57 infants who failed TEOAE but subsequently passed AABR but excluded the 11 infants who passed TEOAE but subsequently failed AABR. The sensitivity and specificity for the community protocol were 80.4% and 99.7% respectively while the positive predictive value was 90.0%. The positive likelihood ratio (PLR) was 268 while the negative likelihood ratio (NLR) was 0.2 in the community. PLR was derived from the ratio: Sensitivity/ (1- Specificity) and NLR from the ratio: (1-Sensitivity)/Specificity [Greenhalgh, 1997]. If our false negative rate of 11/75 were extrapolated for the total population of those who passed TEOAE (n=1,716), the maximum estimate of false negatives would have been 252. The corresponding rates would then be as follows: sensitivity of 15.2%; specificity of 99.7%; PPV of 90.0%; PLR of 50.7 and NLR of 0.8.

4.3. Comparative Performance of the Screening Models

The comparable performance of the hospital- and community-based programmes is outlined in Table 4-9. The 123 babies who passed TEOAE and were scheduled for AABR represent 13.6% (123/902) of all TEOAE passes in the hospital population compared to the 10% (172/1716) in the community. However, because of the different criteria for enlisting subjects for the re-test in the two populations and the logistics of implementing such re-tests in practice particularly in resource-poor settings, this aspect has been excluded from the comparative analysis.

The total number of screening days over a period of 40 weeks in the hospital at a rate of 6 days per week was 240 as opposed to 160 in the community at the rate of 4 days per week. Although the hospital-based programme recorded more screening days, fewer babies (6 versus 13) were screened per day and more babies in the hospital (402 or 30.2% versus 139 or 6.9%) had AABR than in the community-based programme.

The total yield for PCEHL in the hospital was 5.3 per 1,000 (7/1,330) for all categories of hearing loss and 3.1 per 1,000 (4/1,330) for hearing loss ≥40 dB nHL. In contrast, the total yield in the community (taking into account all the infants who failed diagnostic evaluation) was 22.5 per 1,000 (45/2,003) for all categories of hearing loss and 13.5 per 1,000 (27/2,003) for hearing loss ≥40 dB nHL.

Table 4-9. Comparative Analysis of Hospital- and Community-based Screening

Criteria	Hospital Programme	Community Programme
Duration of screening		
• Period	May '05 – Feb '06	July '05 – April '06
• Weeks	40	40
• Days	240	160
No. of babies screened		
• TEOAE	1,330	2,003
• AABR	402 (30.2%)	139 (6.9%)
No. of babies screened- Daily Average [Range]	6 [0 – 13]	13 [1 – 41]
Referral Rates		
• TEOAE	32.2% (428/1,330)	14.3% (287/2,003)
• AABR	10.9% (44/402)	60.0% (82/139)
• Two-stage	3.5% (44/1,274)	4.4% (82/1,855)
Positive Predictive Value	-	90%
Total No. with PCEHL	7	45
Yield per thousand for PCEHL (≥40 dB nHL)	5.3 (3.1)	22.5 (13.5)

The total cost of the hospital programme was US$17,695 (£9,074) compared with US$15,262 (£7,787) for the community programme. Thus, the cost of screening a baby in the community (£3.91) was about half the cost (£6.82) in the hospital (Table 4-10). None of the 44 babies who referred AABR in the hospital SCBU, had previously passed TEOAE. However, in line with the decision to exclude all retest outcomes, the denominator for the AABR

excluded all TEOAE passes to make results comparable with the community outcomes as shown in Table 4-9.

Table 4-10. Cost-Analysis of Hospital- and Community-based Screening

Cost Item	Hospital Programme	Community Programme
	US$	US$
Fixed Costs		
• Equipment [note1]	3,423	3,423
• Staff Remuneration [note 2]	9,115	9,115
Sub-Total	12,538	12,538
Variable Costs		
• Stationery [note 3]	200	300
• TEOAE Tips [note 4]	133	200
• AABR Electrodes, etc [note 5]	4,824	1,668
• Transportation [note 6]	-	556
Sub-Total	5,157	2,724
Total Cost	17,695	15,262
Screening Cost per Baby	US$13.30 [£6.82]	US$7.62 [£3.91]

*See Appendix 4.1 for Notes 1 – 6

4.4. Prevalence/Pattern of Risk Factors for PCEHL

4.4.1. Hospital-based Programme

4.4.1.1. Incidence of Known Risk Factors

The incidence of risk factors for hearing loss in the study population is presented in Table 4-11.

Established risk factors documented in this study were: cranio-facial anomalies, syndromes, prematurity <34 weeks, birth weight <1.5kg, Apgar score 0-4 and 0-6 in 1 and 5 minutes, ototoxic medications and SCBU

admissions. Additional documented risk factors were maternal malaria, maternal HIV and small-for-gestational-age.

Table 4-11. Incidence of Risk Factors among Babies in the Hospital

Risk Factor	No. of Babies	Referred (%)	No with HL (%)
Prenatal			
Family History of Hearing Loss	14	0	0
Maternal HIV	86	10 (11.6)	2 (2.3)
Maternal Malaria	241	14 (5.8)	1 (0.4)
Maternal Rubella	8	0	0
Consanguinity	7	0	0
Congenital Malaria	0	0	0
Craniofacial Anomalies	6	1 (14.3)	1 (16.6)
Syndrome (Downs)	2	1 (50.0)	1 (50.0)
Small-for-Gestational-Age	41	4 (9.8)	1 (2.4)
Postnatal			
Prematurity <34 weeks	39	3 (7.7)	1 (2.6)
Birth Weight <1.5 kg	9	1 (11.1)	1 (11.1)
Apgar Score			
0 - 4 in 1 Minute	404	8 (10.1)	2 (0.5)
0 - 6 in 5 Minutes	285	19 (7.1)	2 (0.7)
Ototoxic Medications	95	5 (6.0)	2 (2.1)
SCBU Admission	180	12 (6.7)	3 (1.7)
Hyperbilirubinaemia and EBT	7	0	0
Bacterial Meningitis	0	0	0
Bacterial Meningitis	0	0	0

Overall, Apgar scores 0-4 at one minute and 0-6 at five minutes and maternal malaria were the most frequent risk factors. However, less than 1% of the babies with such history were confirmed with hearing loss. SCBU admission was documented in the highest number of babies (3/7) with hearing loss. Over 10% of those with history of maternal HIV, craniofacial anomalies, Down's

syndrome and birth weight <1.5kg were referred for diagnostic tests. No baby with a family history of hearing loss failed hearing screening. Similarly, consanguinity was documented in 8 children but none was referred by the hearing screening test. Moreover, none of 7 babies with hyperbilirubinaemia necessitating exchange blood transfusion in the hospital programme failed the screening tests.

4.4.1.2. Characteristics of Children with PCEHL

Multiple risk factors were documented in 4 babies, 3 with severe bilateral hearing loss and 1 with unilateral hearing loss (Table 4-12).

Table 4-12. Risk Factors in Babies with PCEHL Screened in Hospital

Risk Factors	Degree/Type of Hearing Loss					Total
	Profound	Severe	Moderate	Mild	Unilateral	
None	0	0	0	1	0	1
One risk factor	0	0	1	1	0	2
Multiple risk factors	0	3	0	0	1	4
Total	0	3	1	2	1	7

All the 3 babies with severe hearing loss were SCBU graduates and 2 of them received gentamicin for a period of 7 days for neonatal sepsis. Only one baby had no risk factor.

4.4.1.3. Predictors of PCEHL in the Hospital

Predictors of PCEHL among children in the hospital were explored through case-control study of all the children detected with hearing loss matched for age and sex at ratio 1:10 as presented in Table 4-13.

Table 4-13. Predictors of PCEHL among Babies Screened in Hospital

Risk Factors	Cases N=7 (%)	Controls N=70 (%)	Unadjusted OR (95% CI)	p-value	Adjusted* OR (95% CI)	p-value
Prenatal						
Maternal HIV	2 (28.6)	7 (10.0)	3.6 (0.59 – 22.14)	0.145		
Maternal Malaria	1 (14.3)	11 (15.7)	0.89 (0.10 – 8.17)	0.921		
Syndromes	1 (14.3)	0 (0.0)	0.08 (0.04 – 0.17)	0.001		
SGA	1 (14.3)	5 (7.1)	2.17 (0.22 – 21.70)	0.501		
Postnatal						
Prematurity < 34wks	1 (14.3)	0 (0.0)	0.08 (0.04 – 0.17)	0.001		
Apgar Score 1 min						
0-3	3 (42.9)	8 (11.4)	5.81 (1.10 – 30.82)	0.023		
0-4	3 (42.9)	20 (28.6)	1.88 (0.39 – 9.14)	0.431		
Apgar Score 5 min						
0-5	3 (42.9)	7 (10.0)	6.75 (1.25 – 36.52)	0.014		
0-6	3 (42.9)	11 (15.7)	4.02 (0.79 – 20.52)	0.076		
Birth Weight <1.5kg	1 (14.3)	0 (0.0)	0.08 (0.04 – 0.17)	0.001		
SCBU Admission	3 (42.9)	4 (5.7)	12.38 (2.04 – 75.27)	0.001	12.38 (2.04 – 75.27)	0.006
Ototoxic Drugs	2 (28.6)	3 (4.3)	8.93 (1.20 – 66.45)	0.013		

*Adjusted for the effects other variables with values p<0.1 from univariate analysis

Syndromes (p=0.001), prematurity (p=0.001), Apgar score 0-3 in 1 minute (p=0.023) and 0-5 in 5 minutes (p=0.014), birth weight <1,500g (p=0.001), SCBU admission (p=0.001) and ototoxic medication (p=0.013) were significantly associated with hearing loss among the putative risk factors in the univariate analysis. However, SCBU admission emerged as the only risk factor predictive of PCEHL after adjusting for the confounding effects of other variables with p-value <0.1 from the univariate analysis in a multivariate stepwise logistic regression analysis. There was no evidence of any significant interactions among the variables in the logistic regression model.

4.4.2. Community-based Programme

4.4.2.1. Prevalence of Known Risk Factors

The prevalence of risk factors for hearing loss in the study population is presented in Table 4-14.

Table 4-14. Prevalence of Risk Factors among Infants in the Community

Risk Factor	No of Babies	No Referred	No with HL
Prenatal			
Family History of Hearing Loss	9	2 (22.2)	1 (11.1)
Consanguinity	9	0	0
Maternal HIV	n/a	n/a	n/a
Maternal Malaria	395	54 (13.6)	10 (2.5)
Rash in Pregnancy	9	1 (11.1)	0
Congenital Malaria	0	0	0
Craniofacial Anomalies	4	0	0
Syndrome (Downs)	1	1 (100)	1 (100)
Small-for-Gestational-Age (SGA)	83	6 (7.2)	1 (1.2)
Postnatal			
Prematurity <34 weeks	16	5 (31.3)	1
Birth Weight <1.5 kg	9	0	0
Apgar Score	n/a	n/a	n/a
Birth Asphyxia	17	2 (11.8)	1 (5.9)
Ototoxic Medications	151	22 (14.6)	6 (4.0)
Hospital Admission first 28 days	189	31 (16.4)	12 (6.3)
Hyperbilirubinaemia and EBT	32	12 (37.5)	7 (21.8)
Bacterial Meningitis	4	0	0
Delivered Outside Hospital	1,101	156(14.1)	31 (2.8)

n/a: not available

Delivery outside regular hospitals (n=1,101), maternal malaria in pregnancy (n=395), hospital admission (n=189) and ototoxic medications (n=151) were

the most prevalent risk factors. A high proportion of babies with these risk factors except ototoxic medications were confirmed with PCEHL. Over a fifth of those with a positive family history of hearing loss, prematurity and hyperbilirubinaemia requiring exchange blood transfusion were referred for diagnostic evaluation. No baby was documented with consanguinity or congenital malaria. Apgar score was not documented for babies in the community.

Less than one quarter (22.2%) of the babies screened in the community had no identifiable risk factor while majority (57.7%) had only one risk factor. Non-hospital delivery was the most common risk factor documented in 68.9% of the children with hearing loss. It is noteworthy that multiple risk factors were generally associated with hearing loss of at least moderate severity (Table 4-15) although only 20% of those with PCEHL had multiple risk factors.

Table 4-15. Risk Factors in Children with PCEHL Screened in Community

Risk Factors	Degree/Type of Hearing Loss					Total
	Profound	Severe	Moderate	Mild	Unilateral	
No Risk Factor	0	2	5 [1]	2	1	10 [1]
One Risk Factor	0	2	10 [1]	8 [1]	6 [1]	26 [3]
Multiple Risk Factors	0	4 [2]	4 [4]	1	0 [1]	9 [7]
Total	0 [0]	8 [2]	19 [6]	11 [1]	7 [2]	45 [11]

Note: Figures in bracket relate to the false negatives (i.e. infants who passed TEOAE but later confirmed with hearing loss)

4.4.2.3. Predictors of PCEHL in the Community

Predictors of PCEHL among children in the community were also explored through case-control study of all the children detected with hearing loss matched for age and sex at ratio 1:5 as presented in Table 4-16.

Table 4-16. Predictors of PCEHL among Infants in Community Health Centres

Risk Factors	Cases N=56 (%)	Controls N=280 (%)	Unadjusted OR (95% CI)	P-value	Adjusted* OR (95% CI)	p-value
Prenatal						
Family History	1 (1.8)	3 (1.1)	1.68 (0.17 – 16.44)	0.653	-	
Malaria	10 (17.9)	111 (39.6)	0.33 (0.16 – 0.68)	0.002	0.29 (0.13 – 0.61)	0.001
Syndromes	1 (1.8)	0 (0.0)	0.16 (0.13 – 0.21)	0.025	-	
SGA	1 (1.8)	43 (15.4)	0.10 (0.01 – 0.74)	0.006	0.07 (0.01 – 0.55)	0.011
Postnatal						
Non-Hosp Birth	37 (66.1)	147 (52.5)	1.76 (0.97 – 3.21)	0.063	2.48 (1.26 – 4.87)	0.008
Prem < 34 wks	1 (1.8)	2 (0.7)	2.53 (0.23 – 28.36)	0.437	-	
Birth Asphyxia	1 (1.8)	4 (1.4)	1.26 (0.14 – 11.44)	0.084	-	
NNJ + EBT	7 (12.5)	4 (1.4)	9.86 (2.78 – 34.94)	0.000	16.52 (3.75 – 72.71)	0.000
Hosp Admission	12 (21.4)	38 (13.6)	1.74 (0.84 – 3.58)	0.132	-	
Ototoxic Drugs	6 (10.7)	34 (12.1)	0.87 (0.35 – 2.18)	0.763		

*Adjusted for the confounding effects of other variables with values p<0.1 from univariate analysis

In the univariate analysis, maternal malaria (p=0.002), syndromes (p=0.025), Small-for-gestational-age (p=0.006) and hyperbilirubinaemia necessitating exchange blood transfusion (p=0.000) were correlated with PCEHL. Although, non-hospital delivery was not statistically significant in the univariate analysis (p=0.063) it was entered into multivariate stepwise logistic regression model based on the significance level of p<0.1 set as the cut-off rate. The inclusion of this variable was also justified as an index for the diverse adverse obstetric events often associated with births not attended with skilled health workers [WHO, 2004b; Katz, 2003].

After adjusting for the confounding effects of all other variables entered into the model, non-hospital delivery and hyperbilirubinaemia independently emerged as predictors of PCEHL while babies with a positive history of maternal malaria and small-for-gestational-age were significantly less likely to have PCEHL than their controls. There were no significant interactions among the variables in the logistic regression model.

4.5. General Observations and Challenges

4.5.1. Effectiveness of Targeted Screening

Since targeted screening has been mooted as a less costly alternative to UNHS in resource-poor settings [JCIH, 2000; D'Mello, 1995], a retrospective analysis of the effectiveness of selective screening based on the predictor variables for PCEHL in this study population is presented in Table 4-17.

Table 4-17. Targeted Screening based on PCEHL Predictor Variables

Predictor Variable	Hospital		Community	
	Total No.	No. with HL (%)*	Total No.	No. with HL (%)*
Special Care Baby Unit	180	3 (42.9)	n/a	n/a
Non-Hospital Delivery	n/a	n/a	1,101	37 (66.1)
NNJ plus EBT	7	0	32	7 (12.5)
NNJ plus SCBU	203	3 (42.9)	n/a	n/a
NNJ or NHD	n/a	n/a	1,166	42 (75.0)

NHD: Non Hospital Delivery; EBT: Exchange Blood Transfusion; NNJ: Neonatal Jaundice

*Percentage of children with hearing loss; n/a: not applicable

If screening were restricted to SCBU admission/graduates in the hospital-based programme, 42.9% of babies with PCEHL would have been detected. In fact, all the three children with severe hearing loss would have been detected and the yield would not have increased even with the addition of

infants with neonatal jaundice. In contrast, if screening in the community-based programme was targeted at children born outside hospital facilities or with a history of neonatal jaundice about 66% to 75% of children with PCEHL would have been detected. This, of course, is on the assumption that the false negative rate found in this study is similar for both targeted and universal screening.

4.5.2. Follow-Up Default

Overall, 8.2% (273/3,333) of the babies in this study had no conclusive diagnosis because of incomplete tests due to loss to follow-up. Contrary to the 100% uptake recorded in the in-patient screening, follow-up rate was generally less than the recommended target of 70% in spite of the travel support provided to the diagnostic centre. Return for follow-up following active reminders remained poor and well below 20% for the hospital-based study.

A higher proportion (9.0%: 180/2,003) of mothers in the community defaulted compared to 7.0% (93/1,330) of mothers in the hospital-based programme. Although, more mothers in the community defaulted on the 1st stage screening referral, they were significantly more likely to complete diagnostic assessment once referred at the 2nd stage screening compared to mothers in the hospital-based programme (p=0.000).

The documented reasons (Figure 4.17) for follow-up default included, inconsistencies in recorded names and fictitious addresses which resulted in loss to follow-up. Other reasons include: infants sent away to live with grandmothers because of maternal death or special needs, residence outside Lagos, securing husbands' approval, working mother and death of the child.

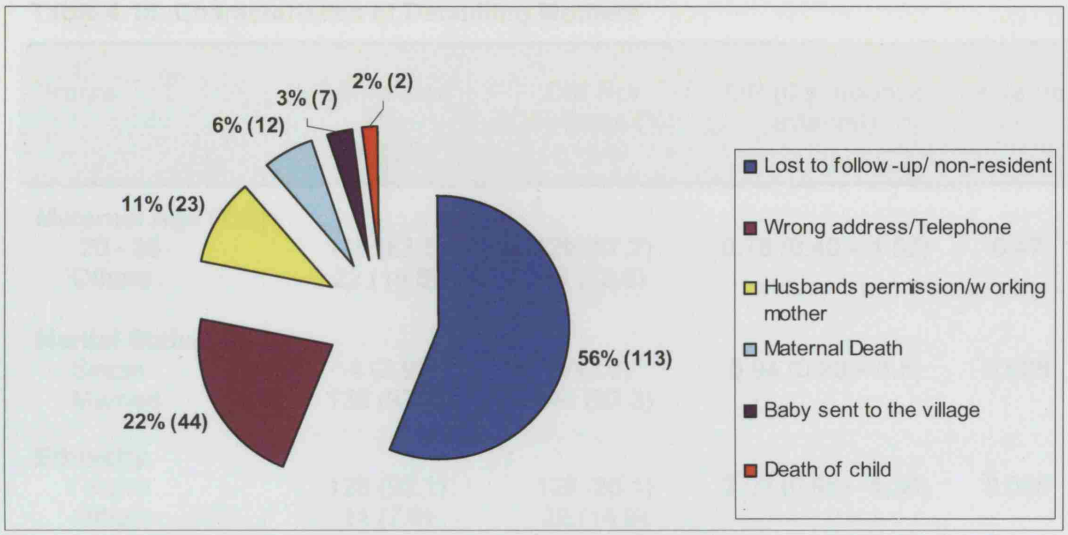


Figure 4.17. Reasons for Follow-Up Default

An exceptional challenge was the very high default rate for follow-up tests scheduled outside routine immunisation visits unlike the regular post-delivery appointment in the hospital. The profile of mothers in the community-based programme who completed the two-stage screening was compared to those who did not complete the required screening after the first stage referral in order to establish the possible reasons for failing to complete the screening protocol (Table 4-18).

Although mothers who did not complete the screening tests were more likely to be single, Christians, multiparous, engaged in small trade or unemployed and not likely to be Yorubas, maternal profile did not show any significant differences between mothers who completed screening and those who did not ($p>0.05$). However, mothers who delivered in non-hospital facilities were significantly more likely to complete the screening tests than mothers who delivered in hospitals ($p=0.034$). In fact, mothers who utilised non-hospital delivery facilities were twice (67.6%) more likely to complete the screening tests than mothers who delivered in hospitals (32.4%).

Table 4-18. Characteristics of Defaulting Mothers

Profile	Completed (%) n=139	Did Not Complete (%) n=148	OR (Confidence Interval)	P-value
Maternal Age (Yrs)				
20 - 35	116 (83.5)	129 (87.2)	0.78 (0.40 – 1.52)	0.47
Others	22 (15.8)	19 (12.8)		
Marital Status				
Single	4 (2.9)	4 (2.7)	0.94 (0.23 – 3.8)	0.928
Married	135 (97.1)	144 (97.3)		
Ethnicity				
Yoruba	128 (92.1)	126 (85.1)	2.03 (0.95 – 4.36)	0.065
Others	11 (7.9)	22 (14.9)		
Religion				
Islam	106 (76.3)	99 (66.9)	1.66 (0.96 – 2.74)	0.058
Christianity	33 (23.7)	49 (33.1)		
Parity				
Primiparous	57 (41.0)	51 (34.5)	1.32 (0.82 – 2.13)	0.253
Multiparous	82 (59.0)	97 (65.5)		
Education				
Secondary & Below	127 (91.4)	128 (86.5)	1.65 (0.77 – 3.52)	0.189
Post Secondary	12 (8.6)	20 (13.5)		
Occupation				
None/Small trade	104 (74.8)	116 (78.4)	0.82 (0.47 – 1.42)	0.476
Full-time job	35 (25.2)	32 (21.6)		
Place of Delivery				
Non-Hospital	94 (67.6)	82 (55.4)	1.68 (1.04 – 2.72)	**0.034**
Hospital	45 (32.4)	66 (44.6)		
Mode of Delivery				
Vertex	134 (96.4)	141 (95.3)	1.33 (0.23 – 2.43)	0.632
Caesarean/Others	5 (3.6)	7 (4.7)		

4.5.3. Excessive Ambient Noise

The maternity hospital was situated on a very busy road. Excessive ambient noise levels from busy traffic and trading activities around the hospital often accounted for widely fluctuating ambient noise levels in the lying-in ward and the SCBU ward. Finding a suitable section within the hospital ward to minimize false-referral rates was a major challenge. A direct consequence of the constantly changing ambient noise levels in the hospital was the

unsatisfactory performance of the Echocheck within the hospital environment. Similarly, the community health centres were also located on major streets with high noise levels from vehicular traffic. Additionally, the immunisation clinics were sometimes difficult to manage when there was a large turnout of mothers, particularly after a holiday. Securing an exclusive use of a quiet room for screening especially in busy clinics also presented a major challenge on a few occasions.

4.5.4. Dealing with Impatient Mothers

Another challenge was mothers' reluctance to wait following hospital discharge when the number of babies awaiting screening was large. And because they lived well outside the hospital's immediate catchments area a significant number never returned after hospital discharge. A similar experience was encountered in the community health centres at very busy clinics when some mothers were required to wait for the hearing screening after their babies had received the vaccination that brought them to the centre. On such occasions, the assistance of the supervising nurses was enlisted to ensure that mothers did not leave before the hearing screening was performed.

4.5.5. Funding Strategy

The screening equipment for this project was donated by their manufacturers. Consumables and all other related operational costs including personnel expenses were financed by a local non-governmental organisation while educational materials were sponsored by a local educational trust. The scope of this project was defined by the available resources. For instance, only two teams of screeners could be afforded for this project which accounted for the 75% coverage of eligible infants attending BCG immunisation clinics in the study location. Similarly, in order to optimise available resources, screening in the community-based programme was initially restricted to one of the four community health centres. However, this strategy had to be abandoned due to the poor uptake in the first two months of the programme. None of the sponsors participated in the design or final report of this project.

Chapter 5

Discussions

5 Discussions

5.1. Rates of PCEHL in Nigeria

A major finding in this research is the high prevalence of PCEHL in our study population against the backdrop of the 6% of infants overall who did not complete the screening protocols. The incidence of PCEHL ≥ 40 dB nHL of 3.1 per 1,000 live births in the hospital is comparable to the reported rates of 1 to 8 per 1,000 from other developing countries [Abdullah et al., 2006; Khandekar et al., 2006; Yee-Arellano, Leal-Garza & Pauli-Muller, 2006; Ali et al., 2000; Chapchap & Segre, 2001]. However, the overall prevalence of 18.9 per 1,000 (35/1,855) infants for PCEHL ≥ 40 dB nHL in the community (pp 167) is only comparable to the 14 per 1,000, reported among newborns in a Jordanian study [Attias et al., 2006] and contrasts sharply with the current global estimate of 6 per 1,000 infants for developing countries [Olusanya & Newton, 2007].

The inclusion of unilateral and mild bilateral hearing loss further raises the incidence of PCEHL in the hospital to 5.5 per 1,000 (7/1,274) and the prevalence in the community to an unprecedented level of 30.2 per 1,000 (56/1,855). This more than five-fold increase between the incidence of PCEHL in the hospital and the prevalence in the community programme perhaps mirrors the often overlooked impact of the prevailing adverse perinatal conditions in developing countries beyond the traditional focus on mortality [Lawn, Cousens & Zupan, 2005]. As previously mentioned in section 2.2.4 (pp 74), both rates of PCEHL in this study are likely to exclude about 23% of children who by 9 months will have hearing loss ≥25 dB HL based on our two-stage screening protocol although majority (>70%) of these children are likely to have mild hearing loss [Johnson et al., 2005]. The incidence or prevalence of PCEHL may also be higher if the study were to be conducted in Northern Nigeria which falls within the meningitis belt and has a high prevalence of consanguineous marriages.

Considering that this study was conducted in the most "developed" urban centre in Nigeria, the prevalence of PCEHL is unlikely to be lower for the rest of the country particularly in the rural areas which account for the vast majority of the populace and have poorer healthcare services. With an annual live births of 5.3 million, the population of infants who are born with or acquire permanent hearing loss in the first three months of life ranges from 26,500 to 148,400 every year. Evidently, PCEHL is a significant public health condition in Nigeria and merits appropriate and timely intervention especially within the context of the millennium development goals on poverty reduction (MDG 1) and universal primary education (MDG 2) [Olusanya, Ruben & Parving, 2006].

5.2. Maternal Profile and Health-Seeking Behaviour

Maternal profile has a great influence on maternal and child health services and it is of special interest in this study because of the sharp contrast between the hospital and community study populations. So also is maternal health-seeking behaviour which may equally reflect individual socio-economic profile. New public health interventions are unlikely to be effective when they fail to take cognizance of prevailing health seeking behaviour in the target population, which most often, is cultural and difficult to alter even with well-planned maternal health education [Pokhrel, 2006; Stekelenburg et al., 2005; Victora et al., 2004a; Bolam et al., 1998a].

While maternal age in this study was normally distributed around the mean ages of 28 and 29 years the very low teenage pregnancy and the older age-group of child-bearing women may reflect the educational status of women in south-western Nigeria in sharp contrast with the pattern in northern Nigeria. It also differed from the profile of women attending immunisation clinics in other developing countries like South Africa, where about one fifth were teenagers, almost 60% were younger than 26 years and majority (82%) were single [Swanepoel, 2005]. Moreover, the total proportion (97.8%) of women

completing primary school education in this study exceeded the national average of 60% [WHO, 2006b].

In addition, women attending hospital-based maternity services in this study were more likely to have tertiary education, than women attending community clinics for BCG vaccination (Table 4-1, pp 143). This finding has been corroborated by other local studies linking the attainment of post-secondary education to uptake of hospital-based maternity services [Onah, Ikeako & Iloabachie, 2006; Ikeako, Onah & Iloabachie, 2006; Osubor, Fatusi & Chiwuzie, 2006]. Two of the reports demonstrated female attainment of post-secondary education, the Christian faith, Yoruba or Ibo ethnicity as significant correlates of uptake of hospital-based maternity services [Onah, Ikeako & Iloabachie, 2006; Ikeako, Onah & Iloabachie, 2006]. Studies in other developing countries have identified maternal educational status as a major determinant of maternal heath seeking behaviour in favour of modern facility-based services [Celik & Hotchkiss, 2000; Bolam et al., 1998b; Raghupathy, 1996; Abbas & Walker, 1986]. This is because education facilitates autonomy of women in making better health decisions. However, it has also been noted that highly educated people visited traditional healers just as frequently as people with less education [Stekelenburg et al., 2005].

Economic empowerment of women, besides formal educational attainment, has been linked to improved socio-economic status and better health seeking behaviour in favour of higher quality heath services [Onah, Ikeako & Iloabachie, 2006; Izugbara & Ukwayi, 2003; Etuk, Itam & Asuquo, 1999; Adetunji, 1992]. In this study, majority of the mothers were employed in some form of small trade or regular jobs. The change from the traditional role of women as housewives to working mothers in the community has significant implications for early childhood care. For instance, women engaged in petty trading were more likely to be within the catchments areas of the screening programme but they were also more likely to default because of the need to earn money for their daily upkeep.

Maternal preferential health-seeking behaviour for non-hospital based maternity services is another notable finding in this study (Table 4-2, pp 146; Figure 4.3, pp 148). This is unexpected in an inner-city area of this commercial and financial capital of Nigeria served by several hospitals within a four-kilometre radius with adequate transport facilities. For most mothers the use of hospital services constituted "alternative medicine", while for a lesser proportion, traditional or unorthodox therapies could be viewed as "alternative medicine". However, births in non-medical institutions, attended by non-skilled personnel, are associated with adverse perinatal conditions that portend great risk for mortality and life-long disabilities for both the mother and the child [WHO, 2004b; Sorensen et al., 2000]. For instance, majority (55%) of mothers in the community delivered in facilities without skilled birth attendants. In effect, the proportion of women with skilled birth attendants (45%) in an inner-city area of Lagos was only slightly higher than the national average of 35% [UNICEF, 2006]. This pattern of health-seeking behaviour which is more often associated with rural dwellers [Osubor, Fatusi & Chiwuzie, 2006; Izugbara & Ukwayi, 2003; Adamu & Salihu, 2002] is in fact not uncommon in major Nigerian urban centres regardless of the availability of public health facilities [Koblinsky et al., 2006; Onah, Ikeako & Iloabachie, 2006; Ikeako, Onah & Iloabachie, 2006; Izugbara & Afangideh, 2005; Akpala, 1998; Nwakoby, 1997].

The predominant reason why mothers used non-hospital based services in the community particularly among the Moslems may be related to their religious beliefs which encourage faith-based services. The reasons why mothers in the hospital-based programme were predominantly Christians could not be ascertained, but it may not be unconnected with the fact that Christianity is largely a product of western civilisation and by implication more tolerant of western lifestyle and medicine. It would therefore appear that the fundamental beliefs of the two religions have some influence on maternal-health seeking behaviour as noted in other studies [Onah, Ikeako & Iloabachie, 2006; Ikeako, Onah & Iloabachie, 2006]. This pattern of health-seeking behaviour is also firmly rooted in superstitious and cultural beliefs in

many developing countries [Stekelengurg et al., 2005; Ezechi et al., 2004; Adamu & Salihu, 2002; Winston & Patel, 1995; Nwakoby, 1994].

Mothers also chose private maternity homes because of the personalised care and emotional support they received from traditional birth attendants during childbirth which is a period of great anxiety for mothers and their families. This support is often lacking in public hospitals where pain management with sub-dural anaesthesia is rare [van Roosmalen et al., 2005]. In fact, some mothers avoid hospital delivery because of their dissatisfaction with certain practices which are considered as disrespectful, inhumane and shameful. For example, offensive language by health personnel or ridiculing of women's poverty, clothing, parity, smell, personal hygiene, cries of pain or desire to remain clothed have been cited as common and undesirable experiences with skilled birth attendants [Koblinsky et al., 2006; D'Ambruoso, Abbey & Hussein, 2005].

A retrospective survey in an urban centre in south-western Nigeria designed to investigate reasons for non-hospital based delivery in women who had attended ante-natal care demonstrated that objection to planned caesarean section and prior caesarean section influenced the preference for non-hospital based maternity services [Ezechi et al., 2004]. While almost half (48.8%) of all deliveries in the hospital in this study were by caesarean section, the majority of births in the community were vaginal deliveries. Hence, mothers who often object to surgical interventions resort to non-hospital delivery to avoid caesarean section or antenatal admission [Ezechi et al., 2004]. The choice of non hospital-based maternity services may therefore emanate from the preferred mode of delivery by mothers offered by the traditional birth attendants. By implication a higher proportion of high risk pregnancies essentially end up with vaginal delivery and its attendant risks.

Further enquiries from mothers using non hospital-based maternity services in our study revealed that the requirement to donate blood, avoidance of caesarean section, better attention and care during the delivery process were

major considerations for their preference. Some mothers also believed that complications during childbirth are due to evil forces, ancestral roots or providence and therefore considered modern obstetric care as ineffective in averting or dealing with these risks. As a result, pregnant women with complications prefer to consult traditional healers even in the face of imminent danger [Okolocha et al., 1998]. Some of these factors could have also influenced the decision by some mothers to deliver at home or in church premises, although the proportion of mothers in either group was about 11% of all those who delivered outside hospital facilities.

The combined use of orthodox and traditional medicine in about 62% of mothers who delivered in hospitals under the community-based programme and who also admitted to using herbal drugs during pregnancy is noteworthy (pp 147). A lower but significant proportion (20%) of mothers in the hospital population also used herbal drugs in pregnancy. This pattern of health-seeking behaviour is equally not uncommon in developed countries such as UK and USA where 20 to 40% of the adult population embrace alternative or complementary medicine for a range of diseases including non-life threatening/chronic conditions [Ernst, 2000; Eisenberg et al., 1993]. The principal motivation among users is that they find alternative medicine to be more congruent with their values, beliefs and philosophical orientation toward health and life and not necessarily (or always) because of dissatisfaction with conventional medicine [Astin, 1998].

Resort to religion and help from prayer houses in addition to the use of herbal drugs, toward commonly perceived spiritual attacks was also not uncommon in our study population as demonstrated in majority (81%) of mothers who delivered in maternity facilities run by churches. Cost did not appear to be a determinant factor since mothers do not pay for antenatal/delivery care in the state hospitals in Lagos except for a token fee of about £20 (N5,000) required for blood investigations and sterile delivery packs. This is consistent with a similar report in another urban area in Northern Nigeria where cost was not found to be a deterrent to hospital deliveries [Ekele & Tunau, 2007]

as would normally be expected to be the case in a developing country particularly among rural dwellers [Borghi et al., 2006; James et al., 2006; Onah, Ikeako & Iloabachie, 2006; Osubor, Fatusi & Chiwuzie, 2006; Palmer et al, 2004; Adamu & Salihu, 2002]. In contrast, mothers pay between £24 (N7,000) to £80 (N20,000) in private hospitals, £12 (N3,000) to £31(N8,000) in traditional maternity homes and up to £21 (N5,300) in church owned facilities where free services have also been reported.

These findings would suggest that the observed maternal health-seeking behaviour in our study is an important consideration that cannot be overlooked for the successful introduction of a "modern" health intervention such as infant hearing screening [Owen, Lewith & Stephens, 2001]. For instance, in our study mothers who delivered outside hospital facilities in the community were significantly more likely to complete the screening protocol which reinforces maternal preference for the combined use of orthodox and traditional health services. In effect, mothers in the community programmes who delivered outside hospital facilities were less likely to default than mothers in the hospital-based programme. This pattern of behaviour could be attributed to the fact that mothers who delivered in hospitals and with professional care were perhaps less likely from anecdotal account, to be concerned about a non-life threatening condition like PCEHL in their children than mothers who used non-hospital based services where the level and quality of newborn care is far below hospital-based practices. More often, the choice of traditional birth attendants in non-hospital facilities is primarily for the mother's safety (due to prevailing superstitious beliefs on the risk of childbirth) rather than the well-being of the newborn in a country where the lifetime risk of maternal mortality (the risk of death during childbirth over a woman's life course) is 1 in 18 [Save The Children, 2007; Ronsmans et al., 2006; UNICEF, 2006]. It is therefore to be expected that such mothers after safe delivery will shift attention from themselves to the well-being of their babies as evidenced by the high-uptake for BCG immunisation shortly after birth.

The high attendance at the community well-child clinic for immunisation may also be attributed to the lack of an alternative traditional therapy for diseases covered under the Expanded Programme on Immunisation (EPI). Consequently, the preference for non-hospital based delivery is unlikely to place an infant hearing screening programme at a greater disadvantage if screening is offered at the community well-child clinics. Besides, the use of non-hospital maternity services by a significant proportion of women in the community further demonstrates the limitation of any maternal and child health intervention in Nigeria that is solely hospital-based, even in urban centres [Harpham & Molyneux, 2001]. The growing support for the integration of traditional birth attendants into mainstream of orthodox maternal health care is in fact likely to make community-based infant hearing screening an appropriate choice in developing countries [Jokhio, Winter & Cheng, 2005; van Roosmalen et al., 2005; Isenalumbe, 1990].

5.3. Feasibility of Infant Hearing Screening Programmes

When the WHA in 1995 urged Member States to make plans for the identification of hearing impairment in babies, infants and toddlers, little evidence on the modalities for achieving this goal existed in many developing countries. Similarly, the Federal Government of Nigeria recognised the need for early detection of childhood hearing loss in the 2005 National Health Policy but the options for pursuing these objectives were uncertain. While hospital-based universal newborn hearing screening was already widely implemented in most developed countries and in a growing number of developing countries, an effective intervention strategy in one country may not always be appropriate in some other countries due to differences in health and socio-economic profile as well as the population health seeking behaviours [Hartley & Wirz, 2002]. This study has demonstrated the value of population-based pilot studies in establishing practical, effective and culturally-appropriate approaches for implementing infant hearing screening programme in a developing country such as Nigeria, particularly among the urban population.

A major finding in this project is that both hospital-based universal newborn hearing screening and community-based universal infant hearing screening programmes are feasible in primary care settings in Lagos, Nigeria using non-specialists without prior audiological expertise. There is no reason to expect that this outcome will be materially different in other urban centres in the country judging by the similarities in maternal health seeking behaviour [Koblinsky et al., 2006; Onah, Ikeako & Iloabachie, 2006; Ikeako, Onah & Iloabachie, 2006; Akpala, 1998; Nwakoby, 1997]. By implication, the early detection of children with PCEHL within an optimal period for effective intervention is achievable in the country against the backdrop of the current delay in detection till as late as an average of 22 months [Olusanya, Luxon & Wirz, 2005a]. Infant hearing screening programmes can thus be readily incorporated into the current WHO initiatives for community ear care training in developing countries as well the current strategic directions for maternal and child health [WHO, 2006a; WHO, 2003]. The excellent collaboration between the research team and the various health administrators in the hospital and community health centres without doubt, significantly contributed to the success of the programme thereby underscoring the value of public-private partnership in the implementation of infant hearing screening programmes in Nigeria.

Volunteer-based community ear care programmes have been successfully implemented in a number of developing countries and may also offer an opportunity for scaling-up infant hearing screening programme systematically through public-private partnerships with the support of all relevant stakeholders [Kennedy, 2004; Hartley & Wirz, 2002; Shrestha, Baral & Weir, 2001]. This is consistent with the recommendations of the WHA 48.9 resolution requesting Member States to facilitate partnership with non-governmental organisations for the effective management of hearing impairment [WHO, 1995]. Finding a suitable testing environment in many primary care settings especially for TEOAE tests which are more susceptible to excessive ambient noise levels may be a major hurdle. Our experience

however, has demonstrated that this challenge is surmountable especially with the support and cooperation from other healthcare professionals.

The prospects for a stand-alone or vertical programme on infant hearing screening are doubtful based on the experience in the first two months of screening in the community. Moreover, a hospital-based programme is inefficient in communities where a significant number of births occur outside hospital facilities. Further studies will be necessary in establishing the generalisability of this outcome to rural parts of the country and to establish peculiar challenges that need to be addressed in these areas. However, it is pertinent to mention that such studies may be hindered in many rural areas by the ethical requirement for the availability of relevant support services before screening is initiated.

5.4. Effectiveness of Infant Hearing Screening

5.4.1. Screening Coverage

The high screening coverage of 97% recorded for the hospital-based programme and the almost 100% in the community-based programme (following screening at the centres where the infants received BCG vaccinations) were well above the 95% JCIH target. Nevertheless, it was difficult to actually determine the true coverage of infants less than 3 months in the BCG clinics against the backdrop of reported lower routine childhood immunisation uptake in some urban areas compared to rural areas [Agarwal, Bhanot & Goindi, 2005; Atkinson & Cheyne, 1994]. The population data of infants under 1 year in 2005 was 9,751 and this figure translates to 4% of total population in the Lagos Island LGA. Based on the 1% of total population estimate by the National Population Commission in Nigeria for babies less than 3 months, the projected population of babies within this cohort in this community programme would have been 2,437 for one year or 2,030 for 10 months. The total number of infants screened of 2,003 (which excludes the 274 babies missed) translates into a screening coverage of 98.6%

(2,003/2,030) which is much higher than the observed proportion of 88% (2,003/2,277).

Two possible reasons can be advanced for the disparity between the total population of infants who visited the BCG clinics during the study period (i. e. 2,277) and the projected population from the census figure for the same period (i.e. 2,030). Firstly, the population of infants less than 3 months may be much higher than the estimated population from the official census figures. Secondly, BCG clinics in the locations used for this project may have attracted many more women outside the LGA as a result of parent to parent communication of this free child care service.

The screening coverage in the hospital was comparable to almost 100% coverage reported in Israel [Attias et al., 2006] and Mexico [Yee-Arellano, Leal-Garza & Pauli-Muller, 2006] and exceeded the coverage of 91% in Brazil [Chapchap & Segre, 2001], 89% in Malaysia [Khairi et al., 2005] and 67% in Oman [Khandekar et al., 2006]. In contrast, the community-based programme performed poorly only for first two months (prior to the subsequent 100% uptake) compared to the 93% coverage in South Africa [Swanepoel, Hugo & Louw, 2006].

Overall, the high screening coverage in this study reflected significant maternal willingness to participate in the new voluntary infant hearing screening programme in this location as no mother withheld consent. This favourable maternal response has been greatly influenced by the inclusion of early hearing detection and intervention in the new National Health Policy and the goodwill from the Federal and State Ministries of Health for the programme. The presentation of the hospital-based programme as part of routine newborn examination with the enthusiastic support of the nursing staff and the presentation of the community-based programme as an essential secondary prevention strategy for PCEHL also contributed to the observed high uptake of this 'new programme'. Additional contributory factors include:

1. The absence of user fees for screening/diagnostic and intervention services.
2. The clear responses recorded for the majority of babies had positive influence on prospective mothers especially at the BCG clinics.
3. Parent-to-parent communication and testimonials of successful screening outcomes in the hospital and also in the community clinics increased the assurance, for prospective mothers, that the screening programme would not cause harm to their babies.

5.4.2. Referral Rates

Of the total population of infants screened under this project 3.8% (126/3,333) were referred for diagnostic evaluation while 6.1% (204/3,333) did not complete the screening protocol. The hospital- and community-based referral rates of the total population of babies for the two-stage screening of 3.5% and 4.4% respectively (adjusting for those who did not complete the screening) and the combined referral rate of 4.0% (126/3,129) were within satisfactory range of the 4.0% target recommended by JCIH. These results would not have been possible without a 2nd stage AABR which for instance accounted for the reduction in the initial referral rate from 27% after TEOAE screening to 3.5% in the hospital-based study; and from 14.3% to 4.4% in the community-based programme. This trend is consistent with the reported outcomes from two-stage screening programmes based on TEOAE and AABR in other countries [Johnson et al., 2005; Lin et al., 2005; Hall 3rd, Smith & Popelka, 2004; Vohr et al., 2001; Gravel et al., 2000; Kennedy et al., 2000; Vohr et al., 1998].

In absolute terms, these rates are also quite satisfactory for an initial pilot programme on infant hearing screening when compared to reported rates from other countries and are likely to improve with time if well managed [Centers for Disease Control and Prevention (CDC), 2003; Watkin, 2003]. For example, in Colorado USA which undoubtedly is one of the leading infant hearing screening locations in the world, the combined referral rate for

hospitals using only OAE screening from 1992 – 1999 was 11.0%; it was 1.5% for hospitals using AABR screening only and 8.4% for hospitals using a two-stage screening with OAE and AABR [Mehl & Thomson, 2002]. In fact, a referral rate of 12.8% was reported in one hospital in Colorado using a two-stage screening protocol. In Rhode Island, USA which pioneered UNHS with current screening technologies in the developed world, the referral rate for an initial TEOAE screening was reported as 10% between 1993 and 1996 [Vohr et al., 1998]. In a more recent report from a large-scale screening programme in Germany the referral rates were 5.2% with TEOAE screening only, 1.9% for AABR only and 2.5% for a two-stage screening [Neumann et al., 2006].

Similarly, when compared with hospital-based newborn hearing screening programmes in other developing countries, the referral rate of 3.5% in the hospital was poorer than the rates of 1.8% reported in Brazil [Chapchap & Segre, 2001] and China [Tang et al., 2006], 1.6% in Malaysia [Abdullah et al., 2006], 1.0% in Israel [Attias et al., 2006] and 0.2% in Mexico [Yee-Arellano, Leal-Garza & Pauli-Muller, 2006] over a screening period of at least one year. Although our referral rate was better than the 10.2% in Pakistan [Ali et al., 2000] and 29% in Philippines [Chong et al., 2003] further reduction is achievable in Nigeria as the programme develops and the screeners become more experienced.

The vast difference between the initial referral rates in the hospital and community was largely due to significant difference in the average age of screening in both settings as discussed in section 5.8.1 (pp 218). It is also pertinent to mention that a further reduction in our referral rates could have been achieved if we excluded unilateral failures in our case definition like the current practice in UK programmes.

5.4.3. Follow-up Return Rate for Diagnosis

The return rate of 61% in the community for diagnostic evaluation was closer to the JCIH target of 70% and exceeded the 11% rate reported in South Africa [Swanepoel, Hugo & Louw, 2006]. Conversely, the return rate of 16%

in the hospital programme was less than the reported rates from similar programmes in other developing countries [Chapchap & Segre, 2001; Tang et al., 2006; Attias et al., 2006; Abdullah et al., 2006; Yee-Arellano, Leal-Garza & Pauli-Muller, 2006]. In-patient screening was completed in the majority of babies in the hospital-based programme. However, because the high-risk screening protocol consisted of both TEOAE and AABR, SCBU babies were more likely to have incomplete evaluation (p=0.000). Subsequent post-natal follow-up after the 2nd-stage screening was generally low in contrast to the much improved return rate in the community. The short follow-up appointment of about one week compared to six weeks in the hospital programme may have accounted for the better follow-up performance in the community.

The very wide catchments areas of the hospital in contrast to the community centres which are designed to serve the population in the immediate neighbourhood could have contributed to the huge loss to follow-up. The few returns from the in-patient screening population were achieved through active and persistent tracking efforts. In contrast, although, the two-stage protocol was incomplete in about 8% of the community group, the better return rate for diagnostic evaluation without any inducement was noteworthy.

Another possible reason for default was that a significant number from the hospital programme (20%) were living outside Lagos metropolis and had left addresses of their relations living in Lagos. Efforts to reach them through these relations were not successful in most cases. For the rest, the reasons for non-attendance varied from the death of the child, difficulty with taking time off work, relocation of mother and child out of Lagos or the baby had been sent to live with grandmother in the village. It is also not unlikely that prevailing superstitious beliefs about childhood hearing loss [Odebiyi & Togonu-Bickersteth, 1987, Ijaduola, 1982] and the predominant preference for traditional medicine may have contributed to some follow-up default. For some mothers, the hidden nature of a potential disability at such an early age may not be sufficiently convincing evidence to warrant the extra efforts to

keep follow-up appointments. A few mothers who were prompted to return for follow-up through personal contacts by the researcher claimed that they forgot the appointments and were perhaps also overwhelmed by the joy of an apparently normal baby.

Follow-up default has also been reported as a major challenge for NHS programmes in developed countries especially in the early stages [Gaffney, 2007; Korres et al., 2006; Mehl & Thomson, 2002; Vohr et al., 1998]. For instance, recent and preliminary data from the Centers for Disease Control (CDC) in USA showed that 68% of infants not passing the screening test did not undergo diagnostic evaluation or have documented evidence of such evaluation [Gaffney, 2007]. The factors adduced for this pattern were similar to those found in this study population with majority (92.5%) falling into the category of unable to contact, "unresponsive" or "unknown".

While lost to follow-up cannot be completely eliminated in our study population, having a dedicated staff or a team of staff (depending on the scope of the screening programme), may be valuable in improving the return rates. For some locations, it may be necessary to have a dedicated vehicle to convey mothers from their homes or more convenient meeting points to the diagnostic centre, which obviously has cost implications for the programme.

5.4.4. Age of Diagnosis

The long delay in returning for follow-up diagnostic evaluation, in the majority (83%) of babies screened as in-patients, increased the age of diagnosis beyond the JCIH recommended target of 3 months. In contrast, the one baby out of the two referred during out-patient screening returned for diagnostic evaluation before the age of 3 months. The mean age of confirmation of hearing loss of almost 8 months (233 days) for babies referred from the hospital-based programme despite active follow-up deserves further investigation as such active follow-up was rarely the case with mothers in the community. In fact, it was not unlikely that some of the parents had started to suspect their babies hearing status by the time they showed up for diagnostic

evaluation as all the 5 babies that so returned were confirmed with hearing loss. The considerable delay by some mothers in the hospital programme may also reflect the fact that they were less likely to be concerned about an invisible non-life threatening health condition in their apparently well children delivered in a specialist maternity hospital compared to mothers in the community-programme who did not have the benefit of skilled birth attendants at delivery outside hospital facilities.

The mean age of diagnosis of about 2 months (58 days) in the community not only fell within the JCIH recommended target but was also comparable to the reported average age of 3 months in Malaysia and Mexico [Abdullah et al., 2006; Yee-Arellano, Leal-Garza & Pauli-Muller, 2006]. Hence, early intervention before 6 months as recommended by JCIH is feasible in the community in spite of the higher average age at screening compared to the hospital-based programme.

Perhaps, and more importantly, the age of identification of PCEHL in this study demonstrates the value of infant hearing screening programme compared to the late detection in the absence of such a programme in Nigeria. Age of diagnosis is generally viewed as a surrogate outcome measure for long-term outcomes such as quality of life, quality of family life, educational and vocational achievement [Davis et al., 1997]. However, it is noteworthy that all the available studies on the optimal period for intervention were conducted in developed countries. Bearing in mind possible differences in phonological structure and trajectory for language development across populations future research will be valuable to ascertain the influences of race and environment on the current thresholds of 6 and 11 months in the USA [Moeller, 2000; Yoshinaga-Itano et al., 1998] or 9 months in the UK [Kennedy et al., 2005 & 2006].

5.4.5. Efficiency of Screening Protocols

The lack of reliable efficiency ratios for the diagnostic tests in the hospital programme precluded a comparative analysis with other studies in which

such data have been reported. In contrast, the sensitivity and specificity reported in the community programme compared favourably with those from studies in the developed world [Kennedy et al., 2005; Hall 3rd, Smith & Popelka, 2004; Watkin, 2003; Vohr et al., 1998].

Similarly, the positive predictive value (PPV) of 90% (45/50) was well within the reported rates of 83.7% from Mexico to 98.2% in Brazil [Khandekar et al., 2006; Yee-Arellano, Leal-Garza & Pauli-Muller, 2006; Habib & Abdelgaffar, 2005; Tang et al., 2006; Chapchap & Segre, 2001]. Comparable rates from UK studies vary from 28% to 67% [Kennedy et al., 2000].

Unlike the PPV, the likelihood ratio (LR) is rarely reported in the literature on infant hearing screening although it has been acknowledged as the best measure of the usefulness of a screening test [Kennedy, 2000; Greenhalgh, 1997]. The positive likelihood ratio (PLR) or negative likelihood ratio (NLR) corresponds with the clinical concept of *ruling-in" or "ruling-out" a disease respectively and incorporates the sensitivity and specificity of a test into a single measure. LR is unaffected by the prevalence of the disease in the population. In this study the PLR of 268 and NLR of 0.2 are comparable with the PLR of 61 and NLR of 0.08 reported by Kennedy et al [2005]. A PLR above 10 is regarded as large and conclusive of the probability of a disease occurring; even as a NLR of <0.1 is large and conclusive of the probability of a disease not occurring [Kennedy, 2000]. The PLR in this study indicates that a positive test is 268 times much more likely to occur in infants with PCEHL than those without while the NLR of 0.2 suggests that the odds of a negative test among infants with PCEHL compared with those without is moderate. However, these results must be taken with caution because of our inability to accurately determine all the true negatives as only 10% of those who passed TEOAE were scheduled for re-testing and only 4.4% were actually re-tested. Extrapolating the false negative rate of 14.7% (11/75) from the random sample for the entire population of infants who passed TEOAE may be more intuitive but the significant reductions in the sensitivity and PLR demonstrated

the limitations of our findings and the need for additional studies against the backdrop of the significant default rates in this population.

5.5. Comparative Analysis of the Screening Models

5.5.1. Babies Screened Daily

The fewer daily average number of babies screened (6) in the hospital compared to 13 in the community programme should not necessarily suggest that hospital-programme was less efficient on this basis. This disparity was principally accounted for by the fewer number of babies available for screening daily in the hospital during the duration of the study which ranged from 0 to 13 babies. The screening team should have been able to handle comparable number of babies daily were they available. The community-based screening has an obvious advantage of attracting more babies than a single hospital or birthing centre because one immunisation clinic serves several birthing centres including non-hospital facilities where infant hearing screening may be impracticable to implement. The wide range of 1 to 41 babies available for screening daily required extra support staff in very busy clinics as a screening staff ideally may only be able to handle 20 infants per day conveniently for TEOAE. Based on the experience in this study, the community-based programme appears to be more efficient in terms of the number of babies screened daily.

5.5.2. Experience with Screening Tests

All measures related to quality control typically found in most screening programmes were implemented to ensure effectiveness. These included adequate supplies of consumables, availability of stand-by/back instruments, overnight charging of screening instruments daily to ensure uninterrupted usage, and several unscheduled visits by the researcher to the screening site to observe the level of cooperation from the mothers and the hospital staff. Since only one person was designated as screener in each of the two teams,

the issue of inter-observer variability was not warranted, more so as the screening instruments were fully automated.

Consequently, our experiences with TEOAE screening (Table 4-6, pp 163) were similar to other NHS programmes in developed and developing countries. However, the test time for AABR was generally much longer than reported in the literature [Meier et al., 2004; Mason & Hermann, 1998]. Test time was longer for babies in the WBN unlike the babies in the SCBU. A possible reason for this disparity may be because of widely practiced exclusive breast feeding at a period when breast milk was not yet established. However, in the community, the practice whereby mothers back their young ones helped to rapidly settle the babies (Appendix 5.1, pp 296).

TEOAE screening was more efficient in the community programme than the hospital programme as TEOAE screening before hospital discharge was associated with a higher referral rate than in the community. About one third (32.6%) of babies screened were referred by TEOAE in the hospital compared to 14.3% in the community. The presence of amniotic fluid/vernix in the ears of some babies past 24 hours after delivery might have accounted for the higher referral rate in the hospital. Further studies are needed to ascertain the cause of this apparent delay in clearing fluids from the babies' ear canals in this study. Overall, AABR screening took much longer than the TEOAE screening and required that the baby was laid in a cot or couch whereas TEOAE screening was conducted with the babies on mothers' laps in the community. However, AABR was necessary to reduce the huge referral rate for follow-up in both programmes. Overall, more babies (37.2%) required AABR in the hospital than in the community (14.3%). By implication, the hospital-based programme was bound to be a more expensive programme.

The South African study [Swanepoel, 2005] is perhaps the only comparable community-based infant hearing screening programme in a developing country using a routine national immunisation platform. However, contrary to

the experience with AABR in the South African study in which considerable difficulty was reported with conducting this test in a community clinic, AABR was successfully conducted among infants attending BCG immunisation clinics in our study. The suggestion by the South African researcher that AABR may not be suitable for community-based screening is not universally valid. The obvious reason for this disparity is due to the choice of the DPT immunisation clinic in South Africa in which the age of the attendances ranged widely from birth to 52 weeks with a mean of 18 weeks. Given the mean age of 2.5 weeks in our study and the fact that children older than 13 weeks were excluded because of the anticipated difficulty with administering AABR to older infants, the experience in South Africa was not unexpected. It is important to note that AABR was equally found to be effective in reducing the overall referral rates in the South African study. The choice of screening protocol for community-based screening must therefore be correlated with the age profile of the target population of infants and the envisaged immunisation platform.

The early age of screening in the hospital-based programme did not translate into early diagnosis within the recommended time limits in majority of babies screened in the hospital compared with the community where most babies who returned for second stage AABR had definitive diagnosis before the age of 5 months. This disparity deserves further investigation as suggested earlier.

5.5.3. Costs per Baby Screened

The costs per baby screened in both programmes were far cheaper than those reported in developed countries. The principal reason for this was the substantially lower staff and operational costs. As in developed countries, it was more expensive to conduct AABR test compared to TEOAE because of the higher costs for consumables.

While expanding the hospital programme beyond the current 6 days to 7 days per week screening would not have made any difference because

babies were not usually discharged on Sundays, the community programme still had two days free. If the 277 babies missed in the community as a result of the initial protocol that required mothers to visit a central location for screening as was experimented in the first two months of this project had been screened, the estimated cost per baby would have reduced from $7.62 [£3.91] to $6.87 [£3.52] as detailed in Appendix 5.2 (pp 297).

In addition, if the estimated 25% of BCG uptake missed by limiting the current programme to four out of the seven immunisation clinics in the LGA was added, an additional 759 babies would have been screened. This additional number could have been accommodated by extending the screening from 4 to 6 days per week as in the hospital, to maintain an average daily screening of 13 babies and cost per baby screened would have been further reduced to $5.49 [£2.82]. This estimate is lower than what has been reported in the literature albeit from hospital-based programmes. For instance, documented cost per baby screened varied from $7.00 in Oman, $9.5 in Mexico, $16.00 in Taiwan to £14.00 or $27.00 in the UK [Khandekar et al., 2006; Yee-Arellano, Leal-Garza & Pauli-Muller, 2006; Lin et al., 2002; Kennedy et al., 2000].

Overall, community-based UNHS was more cost-effective and more efficient in Lagos than hospital-based UNHS. However, some limitations in our cost analysis should be noted. For instance, indirect costs related to the provision of facilities at the screening sites, the time spent by hospital and community staff in educating mothers as well as the complete cost of identifying a child with hearing loss was not considered. Moreover, there were few occasions where a child was tested more than once with TEOAE or AABR but such additional costs were minimal and could not have materially affected our results. The implicit and explicit costs associated with false negatives, false positive referrals and extra efforts to increase coverage may also have impact on the performance of the screening models as noted by Davis et al [2001]. However, these costs were not considered due to the lack of relevant data and there is no evidence to suggest that they would have materially

altered the relative efficiency of community-based over hospital-based programme. For instance, the costs associated with the intensive follow-up efforts for mothers referred under the hospital-based programme would have made that screening model more expensive than reported in this study. In addition, any comparative analysis of the cost per infant detected with PCEHL would have been distorted by the substantial disparity in the default rates between both programmes.

5.5.4. Yield for PCEHL

The yield of 3 per 1,000 (4/1,330) for PCEHL in the hospital-based study for hearing loss ≥ 40dB nHL is more than double the UK rate of 1.2 per 1,000. However, as in the UK study, the yield in this study nearly doubles with the inclusion of mild and unilateral hearing losses to 5.3 (7/1,330) [Wakin, 2003]. The more-than four-fold yield of 22.5 per 1,000 (45/2,003) for PCEHL in the community-based programme and the almost 50% reduction in screening cost per child evidently makes this model the preferred option in this population. In fact, the much higher yield in the community would seem to justify the higher AABR referral rate in the community from an economic standpoint. The significantly higher proportion of infants with acquired early-onset hearing loss from neonatal jaundice requiring exchange blood transfusion, neonatal sepsis, ototoxic medication and hospital admission in the neonatal period in babies born outside hospital facilities, may account for the higher yield for PCEHL in the community. This figure may be indicative of a true increase in the prevalence of hearing loss within the first 3 months of life.

5.6. Evaluation of Risk Factors for PCEHL

The knowledge of the risk factors for PCEHL in any population has two major advantages. Firstly, it helps in directing efforts towards primary prevention for PCEHL. Secondly, it is valuable in determining relevant variables or conditions for a targeted screening programme which may be an interim strategy before the introduction of UNHS in some settings. For these

reasons, studies on infant hearing screening in many countries have also focused on the identification of predictors for PCEHL besides other outcomes [Uus & Bamford, 2006; Kountakis et al., 2002; Vohr et al., 2000; Davis et al., 1997; D'Mello, 1995].

5.6.1. Significant Risk Factors for PCEHL in Nigeria

Of all the risk factors considered in the hospital population, admission into the SCBU was the only predictor of PCEHL. Furthermore, SCBU babies were also more likely to have PCEHL as they were often products of difficult deliveries from prolonged obstructed labour, prematurity, breech delivery and vacuum extraction. In this study a significant number of mothers (about 35%) who delivered in the hospital were unbooked and referred for emergency caesarean section from centres with limited facilities. Delays in receiving prompt attention due to theatre related factors such as irregular power supply, ongoing emergency surgery and shortage of anaesthetists have been reported [Ameh, Dogo & Nmadu, 2001]. Overall, in the hospital population, 65.0% (117/180) of SCBU admissions, were delivered by caesarean section as against 39.8% (458/1,150) of babies in the WBN (OR: 2.81; 95% CI: 2.00-3.95; p=0.000) as shown in Table 4-3 (pp 149). Furthermore 20.3% (117/575) of babies delivered by caesarean section were admitted into SCBU compared to 6.8% (47/696) of normal deliveries. It is worth noting also that facilities in the SCBU lacked intensive care and were far less sophisticated than those typically found in NICU in developed countries. So even with the best of skills, the quality of care is often sub-optimal because of these constraints [Ameh, Dogo & Nmadu, 2001]. These constraints are also more likely to be compounded at the secondary or community level of care [Garg, Krishak & Shukla, 2005].

A notable finding in the community-based programme was the significant association between neonatal jaundice (NNJ) and PCEHL (Table 4-16, pp 177). This is consistent with similar findings in other developing countries like Jordan and Malaysia [Attias et al., 2006; Mukari, Tan & Abdullah, 2006; Boo et al., 1994]. NNJ is the most common condition requiring evaluation and

hospital admission in the first week of life [Kilic, et al., 2005; Owa & Osinaike, 1998] and constitutes a significant cause of neonatal mortality and morbidity [Gordon et al., 2005; Ezeaka et al., 2004; English et al., 2003] and PCEHL [Boo et al., 1994]. In fact, NNJ has been reported as the highest contributor to neonatal mortality [Kilic et al., 2005; Owa & Osinaike, 1998]. Because of the high prevalence of NNJ and the associated discolouration of the skin, sclera, and mucus membranes, it was readily and reliably elicited from mothers at the immunisation clinics regardless of where the babies were born. Unfortunately, NNJ is rarely recognised in global burden of disease estimates as a significant cause of infant mortality, morbidity and disability in the developing world [Lawn et al. 2007, Lawn, Cousens & Zupan, 2005; Bryce et al., 2005].

Icterus neonatorium or physiological jaundice at serum levels not exceeding 5-6 mg/dl (86-103μmol/l) is a benign transitional phenomenon in the vast majority. However, in some babies, serum bilirubin may rise to hazardous levels capable of causing brain damage. The resulting bilirubin encephalopathy which has been linked with the deposition of unconjugated bilirubin in the cochlear nuclei and the yellowish staining of the basal ganglia [Newton, 2001] may lead to kernicterus, a devastating clinical tetrad of choreoathetoid cerebral palsy, sensorineural hearing loss, palsy of vertical gaze and enamel hypoplasia of the deciduous teeth. Hyperbilirubinaemia has been identified as a significant aetiological factor among school children that are deaf in West Africa with a prevalence rate of 1.9 to 5.7% [Olusanya & Okolo, 2006; Brobby, 1988; Ijaduola, 1982].

G6PD deficiency (a mutation located on the X-chromosome) is the commonest enzyme disorder of human beings and a globally important cause of neonatal jaundice. Because G6PD deficiency is X-linked, it is more likely to affect males than females who must have two defective copies of the gene to manifest the disease. The significantly higher prevalence of PCEHL among males in our community-based study may in fact mirror the causal relationship between PCEHL and the X-linked G6PD deficiency. The G6PD

enzyme catalyzes the oxidation of glucose-6-phosphate to 6-phosphogluconate and also reduces the oxidized form of nicotinamide adenine dinucleotide phosphate ($NADP^+$) to nicotinamide adenine dinucleotide dehydrogenase phosphate (NADPH). It protects the red cell membrane against oxidative stresses by maintaining glutathione in its reduced state. G6PD deficiency is asymptomatic in the vast majority while the most common clinical features in the few that are symptomatic are neonatal jaundice and acute haemolytic anaemia.

G6PD deficiency affects all races. The highest prevalence has been reported from Africa, Asia, and the Middle East. G6PD deficiency has been classified into three types: low, normal, or increased activity levels of the enzyme. A variant, G6PD-A, encountered in African Americans, has been reported as the predominant group [Kaplan et al., 2004; Slusher et al., 1995]. Severe deficiency variants primarily occur in the Mediterranean population while the enzymatic variants in the African population have more activity and produce a milder form of the disease.

Apart from the association with hyperbilirubinaemia, G6PD like the abnormal haemoglobins is protective against falciparum malaria and as many as 22% of Nigerians may be G6PD deficient while up to 17.6% preterm infants may be affected [Owa & Dawodu, 1990]. Exposure to haemolytic agents such as menthol and/or camphor containing domestic products (like mentholated powder used for cord dressing) and the resultant unconjugated hyperbilirubinaemia from rapid haemolysis of the red cells have been implicated in the pathogenesis of this condition in Nigeria [Olowe & Ransome-Kuti, 1981]. G6PD deficiency has also been reported as the most important aetiological factor for hyperbilirubinaemia in Nigerian babies, accounting for about 60% of neonatal jaundice [Olowe & Ransome-Kuti, 1981].

The introduction of anti-D factor, phototherapy and double volume exchange blood transfusion in the treatment of unconjugated hyperbilirubinaemia has

significant impact on the incidence of kernicterus. Researchers have demonstrated that prompt treatment with phototherapy and exchange blood transfusion, are effective in reversing the neurotoxic effects of unconjugated hyperbilirubinaemia [Wong, Chen & Wong, 2006; Deorari et al., 1994]. In this study, all 7 babies with documented hyperbilirubinaemia who also had exchange blood transfusion in the hospital group passed hearing screening in sharp contrast to the outcomes in the community group. The lack of association between hyperbilirubinaemia and PCEHL in the hospital may be related to appropriate monitoring and prompt management of neonatal jaundice in babies on admission in contrast to the community where mothers' health seeking behaviour in favour of traditional medicine cause considerable delay with presentation to hospital.

In addition, the lack of SCBU/NICU in most private hospitals and health centres also leads to delay in carrying out exchange blood transfusion from late referral to secondary or tertiary hospitals. Community workers have also been reported with significant knowledge gaps about the pathophysiology of neonatal jaundice [Ogunfowora & Daniel, 2006]. For instance, a significant number of health workers believe in the efficacy of herbal drugs and most of them would treat neonatal jaundice with antibiotics and exposure to natural light before referral to hospital. This gap has also been advanced as contributing to the late presentation of babies with neonatal jaundice to hospitals as hospital visit is often delayed until kernicterus becomes evident [Ogunfowora & Daniel, 2006].

The WHO working group [WHO, 1989] recommended population screening of all newborn babies in areas with a prevalence rate of 3-5% G6PD or more in males for early identification and management of pathologic hyperbilirubinaemia. Recently, emergency treatment for neonatal jaundice has also been recommended as part of the measures for reducing neonatal mortality and for achieving MDG 4 that targets two-thirds reduction in child mortality rate by year 2015 [Adam et al., 2006]. The findings in this study underscore the need to implement the recommendations of the WHO

working group towards the reduction and effective management of the burden neonatal jaundice in Nigeria and other developing countries.

Another major and unique finding in this study is the association between non-hospital birth and PCEHL (Table 4-16, pp 177). This finding clearly suggests that the risks attached to births without skilled attendants in developing countries are real and extend beyond maternal and infant mortality. There is currently no evidence in the literature on the pattern or causal pathway from the obstetric experiences of babies with PCEHL born outside hospital facilities. This could be attributed to the fact that such occurrence is limited to developing countries and would therefore merit further investigation. Current maternal education should incorporate this additional risk associated with non-hospital based deliveries.

Of special interest is the value that prospective mothers attach to vaginal delivery which is a major motivation for seeking non-hospital based delivery - an important predictor of PCEHL in the community. However, this mode of delivery portends significant risks to both mother and child [Ezechi et al., 2004; Onah, Ikeako & Iloabachie, 2006]. Traditional birth attendants often pride themselves in their ability to deliver every baby vaginally irrespective of the risks to the babies. As observed in one of the studies, mothers attending antenatal care in a tertiary hospital [Ezechi et al., 2004], opted for non-hospital services, unaware of its attendant risks, to avoid caesarean section or ante-natal admission. It is not unlikely that mothers in this study held similar views as almost all of them attended antenatal clinic.

Caesarean section is not culturally acceptable to mothers who consider non-vaginal delivery as a sign of maternal laziness or a curse from perceived enemies or deity. Unavoidable surgical intrapartum intervention therefore occurs late and only where there is a glaring failure of vaginal delivery. Unknown to the parents this disposition often compromises the quality of such babies who would have suffered significant damage from preventable intrapartum stress. For instance, in another tertiary hospital, in the southwest,

trial of labour for second delivery following caesarean section was documented in 67.6% of the mothers with previous caesarean section. Trial of labour in the study, ended with emergency caesarean section in over half of them [Nwokoro et al., 2003]. However, for those who have no facilities for surgical intervention, vaginal option may result in grave consequences for the child even if the mother is spared. Unfortunately conditions such as PCEHL and other developmental problems are hidden and by the time they evolve, they may not be readily linked to the events that surrounded the birth.

Antenatal clinics may provide an effective forum for educating mothers on major issues of maternal health and early childhood development as majority of mothers in our study population attended antenatal clinic. Similarly, the culturally held belief which favours only vaginal delivery portends great danger for the high-risk unborn child. Health education, during antenatal clinics therefore, needs to be culturally relevant to cause a significant change in behaviour that will have considerable impact on early childhood outcomes.

In the same way, health education on the avoidance of haemolytic agents like camphor, which are commonly used as insect repellent or for the manufacture of mentholated and herbal preparations, may be helpful in curtailing this risk of neonatal jaundice in many households. In addition, mothers need to be educated on the early warning signs and the dangers that late presentation holds for the long-term speech, language, cognitive and motor developmental outcomes in the affected children.

In this study population small-for-gestational-age babies were significantly less likely to have PCEHL (Table 4-16, pp 177). This finding is not fully in agreement with other reports among small-for-gestational-age infants. For instance, one study showed no significant difference in the incidence of PCEHL among small-for-gestational-age survivors and their appropriate-for-gestational-age twin pairs [Monset-Couchard, de Bethmann & Relier, 2004]. On the contrary, another study demonstrated a significant increase in the incidence of PCEHL among extremely preterm small-for-gestational-age <28

weeks compared to their appropriate-for-gestational-age controls [Bardin, Piuze & Papageorgiou, 2004]. Whereas the risk for PCEHL was significantly higher in preterm small-for-gestational-age infants than their controls, full term small-for-gestational-age infants were either at the same level of risk or lower. Notwithstanding, various reports have shown reasonable agreement in similarity for other neurodevelopmental problems in both classes of small-for-gestational-age infants compared to their controls [Monset-Couchard, de Bethmann & Relier, 2004; Das & Sysyn, 2004; Bardin, Piuze & Papageorgiou, 2004; Larroque et al., 2001]. Documented neurodevelopmental problems include: behavioural and speech problems, proportionately small size at 3 years, severe handicaps, cerebral palsy, blindness, mental/motor deficiencies, late entry into secondary school and poorer school performance.

The aetiology of small-for-gestational-age is multifactorial and complex involving both genetic and non-genetic factors. Non-genetic factors are related to the socio-economic and health status of mothers such as maternal weight gain during pregnancy, haemoglobin level at booking, gestational age and intra-uterine infections. Causes of diminished foetal growth are primarily hypoxia and genetic disorders. Although small-for-gestational-age infants in the long term have been shown to have increased rates of neurodevelopmental disorders and behavioural disturbances, the ensuing alteration in foetal nutrition leads to significant adaptation for survival in the short-term. Besides, women who were born small-for-gestational-age have been shown to have increased risk of giving birth to small-for-gestational-age infants [Das & Sysyn, 2004]. The lower risk of PCEHL in small-for-gestational-age infants in this population may therefore result from the protective short-term adaptation to nutritional deficiency, having small-for-gestational-age mother or genetic factors.

In this study, infants with a positive history of maternal malaria were less likely to have PCEHL (Table 4-16, pp 177). However, maternal malaria was not verified and it is quite possible that mothers' positive history of malaria

was a wrong diagnosis of other febrile illnesses. Notwithstanding, Nigeria is a holoendemic region for malaria and repeated infections are common and stable. Since majority of the population is immuned, epidemics do not occur. Although, the ability to limit the rate of parasitaemia is restricted and may lead to extensive haemolysis and anaemia in pregnancy, majority of women in this study attended ante natal clinics where anti-malaria, folic acid and multivitamin tablets were given routinely.

Family history did not show any statistical association in this study possibly because of the very small numbers of families reporting positive history. Cultural beliefs in the southwest makes it difficult for family members to admit to a positive history of disease as this admission may portend significant social stigma/isolation for affected individuals and their families. This finding contrasts sharply with the finding by Swanepoel [2005] in which family history of hearing loss was documented in a significant proportion of infants attending immunisation clinics. In addition, the high rate of non-disclosure of maternal HIV status as reported in the South African study seems to justify the exclusion of this information in our community-based study.

Contrary to other developing countries and the influence of Islamic religion in this study population, consanguinity was rare because of the strong cultural belief of the predominant ethnic group which forbids consanguineous marriages. The low prevalence of consanguinity may also account for the very low positive family history obtained in this study apart from the cultural practice of non-disclosure of disabilities.

Although low Apgar scores at 1 or 5 minutes were associated with hearing loss in the univariate analysis, they were not predictive of PCEHL in our hospital population after adjusting for possible confounders in the multivariate logistic regression analysis. This pattern was similarly suggested by Mencher & Mencher [1999]. While some researchers have low Apgar scores as risk factors for PCEHL [Vohr et al., 2000; Eichwald & Mahoney, 1993] others have not [Khairi et al., 2005; Kountakis et al., 2002]. In view of the relative

ease of obtaining this routine clinical information in hospital deliveries and the mixed reports on the statistical association between Apgar scores and PCEHL further studies may be worthwhile in establishing its clinical usefulness as a possible pre-screening tool in Nigeria.

It is not known whether the observed risk factors for PCEHL found in this study will be significantly different in other parts of the country. For instance, consanguineous marriages may emerge as additional risk factor in the northern parts of the country. Moreover, SCBU admission may be irrelevant for the rural population not served by regular hospitals with such facilities.

5.6.2. Prospects for Targeted Hearing Screening

About 25% (14/56) of the babies with hearing loss in the community and only 14% (1/7) of those confirmed with hearing loss in the hospital, had no identifiable risk factor(s). The higher prevalence of risk factors in this study reflects the relatively higher acquired adverse perinatal conditions in our study population. However, because of the limitations of risk factors as prognostic tools even when several independent risk factors are combined [Ware, 2006; Wald, Morris & Rish, 2005; Watkin et al., 2005], targeted screening based on these risk factors should not be regarded as an alternative to universal infant hearing screening. In fact, as Wald [2001] rightly noted, the process of identifying potential risk factors itself makes targeted screening "universal". Moreover, the preselection of risk factors to be considered in this study based on the JCIH list would have foreclosed the identification of other potential and previously unknown variables that could be additional predictors of PCEHL in this population as demonstrated in this study with non-hospital delivery. It is axiomatic that an initial pilot UNHS programme is required to identify the relevant risk factors for an effective targeted screening in any community.

Targeted infant hearing screening has been recommended as a cost-effective strategy for developing countries and may intuitively appear desirable in this study population [JCIH, 2000; Mencher & Mencher, 1999;

Gell et al., 1992]. For instance, only 180 SCBU babies would have been screened in the hospital bringing down the overall cost of screening from US$17,695 (£9,074) as in Table 4-10 to US$15,463 (£7,930) as shown in Appendix 5.3 (pp 298). However, due to the impact of fixed costs (screening equipment and personnel), screening cost per baby would have risen from US$13.30 (£6.82) to US$85.91 (£44.05). Unfortunately, this factor is rarely reflected in the arguments in favour of targeted screening even for developed countries [Kileny & Lesperance, 2001; Paradise, 1999; Curnock, 1993] and is unlikely to be mitigated by the identification of additional risk factors not currently listed by JCIH [Kountakis et al., 2002]. However, given the small number of high-risk babies, it is possible to consider entrusting screening to the existing nursing staff in the hospital thereby eliminating separate personnel costs. Accordingly, the total cost of screening would drop sharply from US$15,463 (£7,930) to US$6,348 (£3,255) which translates to US$35.27 (£18.09) per baby screened.

In contrast, if targeted screening was introduced for our community-based programme the overall cost would have doubled from US$15,262 (£7,827) to US$31,485 (£16,146) while the cost per baby screened also would have increased more than three-fold from US$7.62 (£3.91) to US$27 (£13.85) as shown in Appendix 5.3 (pp 298). This is principally because all the "high risk" infants born outside hospitals or with a history of neonatal jaundice would have had to undergo AABR screening with the attendant cost implications. Although all babies with auditory neuropathy in the high risk group would have been detected by such a protocol, infants with auditory neuropathy and PCEHL in the non-risk group would have been missed. In practice, the actual coverage from a prospective targeted hearing screening in an environment strongly associated with unfavourable superstitious beliefs and attitude towards hearing impairment is likely to fall significantly below the 75% from our retrospective analysis. This is partly because mothers may be unwilling to volunteer information on these risk factors for screening purposes as it may unduly stigmatise or label their babies. Moreover, the logistic and the

administrative costs of implementing this protocol in a busy immunisation clinic will significantly undermine its effectiveness in this target population.

5.7. Referral for Intervention Services

All mothers whose babies were referred for early intervention services were counselled on the importance of effective communication through multimedia presentations and video clips. A typical counselling session with mothers of infants with PCEHL is shown in Appendix 5.4. Infants with severe-to-profound hearing loss were referred for specialised intervention services including the fitting of appropriate hearing aids. In this study, infants with mild-to-moderate hearing loss were enrolled in a surveillance programme because of the difficulty of convincing parents of the need for hearing aids for these levels of hearing loss at this early stage. Some reports have suggested that even in the absence of definite therapeutic treatment such as amplification devices early detection in general provides enormous psychological and emotional benefits to children and their parents [Bailey, Skinner & Warren, 2005; Love et al, 2005; First & Palfrey, 1994].

Consequently, the importance of managing the child's environment for effective communication, to enhance signal-to-noise (S/N) ratio, was highlighted especially for babies with minimal (mild bilateral or unilateral) hearing loss. Mothers were educated on the possible difficulties and fatigue from increased listening efforts such children may encounter in a noisy environment or when they are far from the source of sound [Hicks & Tharpe, 2002]. Those with unilateral hearing loss may experience difficulties with localising distant sound [Dodd-Murphy & Parker, 1998; Newton, 1983]. Because of the possible impact of these categories of hearing loss on speech clarity and understanding, mothers were encouraged to engage the child's attention while maintaining eye contact when talking to the child. The importance of speaking clearly under good lighting and especially to the good ear for effective communication was also demonstrated. The need to monitor

the evolving speech and language development and seek help when they have concerns about their child's development was equally stressed.

Another important component of parental counselling was to raise awareness on the long-term adverse impact of possible child abuse or neglect as a result of the child's difficulties with communication in early childhood as well as the risks associated with traditional and unorthodox therapies for deafness [Andrade & Ross, 2005; Knutson, Johnson & Sullivan, 2004; Lasisi & Ajuwon, 2002; Henggeler et al., 1990; Quittner, Gluekauf & Jackson, 1990; Odebiyi & Togonu-Bickersteth, 1987; Togonu-Bickersteth & Odebiyi, 1985]. The significance of parents' ability to accurately recognise, understand and interpret their children's behaviour, body language, facial expressions and other communicative signals in fostering parent-child interaction and attachment and improved social-emotional development was emphasised [Howe, 2006; Koester & Meadow-Orlans, 2004; Vaccari & Marschark, 1997].

It was not possible within the scope of this study to establish the possible impact of the strong preference for combination therapy in this population on the uptake of modern intervention services such as the use of amplification devices in infants. This may merit further research in view of the stigma often associated with such devices which may result in a significant number of children with hearing aids not using them [Mukari et al., 1999]. Nonetheless, early detection provides numerous benefits beyond early intervention with amplification devices which are important for the infants and the parents in a developing country like Nigeria as enumerated in section 2.4.4 (pp 101 – 105). For instance, where the provision of amplification devices backed up with appropriate support services, early enrolment in sign language classes is encouraged to provide a communication mode between the hearing-impaired child and their parents as soon as practicable. This will be a significant achievement considering the current substantial delays in the enrolment of such children in the schools for the deaf in Nigeria and the benefits of such early intervention [Olusanya, Luxon & Wirz, 2005a; Meadow-Orlans et al., 2004; Hindley & Parkes, 1999].

It is pertinent to mention that the service delivery strategy under this pilot project where screening services were "community-based" whereas diagnostic and other intervention services were provided at a specialist institution outside the immediate screening sites may account for low uptake of intervention services. In order to optimise uptake of these services as resources become available and the infant hearing screening programme is taking to scale, it would be necessary to systematically move away from institutional to community-based rehabilitation in line with the recommended approach for the management of individuals with disabilities in developing countries [WHO, 2004c; Hartley & Wirz, 2002; Turmusani, Vreede & Wirz, 2002].

5.8. Further Observations and Challenges of UNHS

5.8.1. Test Environment for Hearing Screening

Maintaining acceptably low referral rates in TEOAE-based NHS programmes has been linked to the environment, the equipment and the infant. Achieving a quiet environment, a good probe fit and a quiet baby then leaves the infant's age of screening as a necessary determinant factor for a clear response. For instance, a study in Taiwan documented a drop in the referral rate from 22.4 at birth to 6.4% by the age of 72 hours before hospital discharge [Lin et al., 2002]. The value of reducing referral rate with increasing age is corroborated in that study where the referral rate following TEOAE in the community was 14.3% compared to 32.6% reported for the younger hospital population. However, with a predominant older population (>72 hrs), the 14.3% referral rate in this study is far in excess of the 6.4% from Taiwan. Against the backdrop of greater TEOAEs which have been postulated in the Negroid race [personal communication: Professor David Kemp, June 1, 2007], the generally recorded high ambient noise levels at the screening sites emerged as the most suitable explanation for the high referral rate.

The ambient noise levels of 59.3 to 90.5 dBA, documented from the screening sites were far in excess of the reported 30 to 59 dBA in the UK

[Hunter et al., 1994]. Even in the so-called quiet rooms the noise levels were in excess of 60 dBA ranging from 60.3 to 67.9 dBA compared to a maximum level of 36 dBA in the UK [Hunter et al., 1994]. Additionally, the widely fluctuating noise levels in these quiet rooms might have contributed to the high referral rates in this study. For instance, the AABR referral rates of 3.5 and 4.4% exceeded the rates in other studies such as 0.2 in Mexico, 1.04 in Israel, 1.8 in Brazil and 1.4 in the UK [Yee-Arellano, Leal-Garza & Pauli-Muller, 2006; Attias et al., 2006; Chapchap & Segre 2001; Kennedy et al., 2005]. In contrast, because of the two-stage protocol combining TEOAE with AABR, the rates in this study were better than the 9 to 14.7% reported from Taiwan, Pakistan, South Africa and Malaysia, [Ali et al., 2000; Lin et al., 2003; Abdullah et al., 2006; Swanepoel, Hugo & Louw, 2006].

The recorded high ambient noise levels in both the hospital- and community-based settings is therefore an indication of the significantly high noise levels in this inner city area of Lagos. It is therefore logical that the Echocheck was unable to perform satisfactorily judging from the reported lower ambient noise levels in the UK or in the many countries worldwide where this model is used for newborn hearing screening. This finding therefore underscores the crucial role for equipment trial runs to verify their suitability for a particular environment, before embarking on any screening programme. However, our experiences with the Echocheck cannot be generalised for the rest of the country. Hospitals outside the main large cities may not have as much difficulties with ambient noise. Moreover, it is not unlikely that newer models of Echocheck may have more robust noise tolerance limits to enhance their wider applicability in developing countries than the older models.

5.8.2. False Negative Results

Also of great interest and public health significance is the false negative rate of 0.6% (11/1,716) in this study. This rate (which is likely understated because of the high follow-up default) is comparable to the reported rate of 0.2% in Jordan but much higher than the rate of 0.01% from the UK. About two thirds (7/11) of the babies with false negative result had neonatal

jaundice and 43% (3/7) of them had exchange blood transfusion. The degree of hearing loss was severe in 2, moderate in 6, mild in 1 and unilateral in 2. The two babies with severe hearing loss also had double volume exchange blood transfusion.

The high incidence of false negative results reported in babies with severe hyperbilirubinaemia is an indication that the overall prevalence of PCEHL in this study may be an underestimation. In addition, false negative results have significant implications for co-existing auditory neuropathy or hyperbilirubinaemia-induced auditory dysfunction [Shapiro, 2003]. The term "auditory neuropathy" was originally coined in 1996 after the first longitudinal study of 10 subjects who were discovered to have absent or grossly abnormal auditory brainstem responses in the presence of normal otoacoustic emissions [Starr et al., 1996]. Auditory neuropathy is related to conditions which selectively affect the spiral ganglion cells, their axons or the auditory nerve while excluding pathologies which affect the central auditory pathway from the brain stem as far as the cortices [Rapin & Gravel, 2003]. However, excessive exposure to free unconjugated bilirubin has been associated with a variety of neurological sequelae, including auditory neuropathy and other central auditory processing disorders originating from lesions in the dorsal and ventral cochlear nuclei, superior olivary complex, nuclei of the lateral leminiscus, inferior colliculus, globus pallidum and subthalamus [Bamiou, Musiek & Luxon, 2001; Starr et al., 1996]. The resultant dys-synchrony in the auditory pathway has been linked with attention deficit hyperactivity disorder (ADHD), learning disability and developmental dyslexia with significant implications for future learning. Moreover, an association between moderate-to-severe hearing loss and central auditory dysfunction has also been established with hyperbilirubinaemia in the absence of kernicterus [Shapiro, 2003].

The prevalence of auditory neuropathy in newborns ranges from 2.2% to 17.3% and may be as high as 24.1% among NICU babies [Ngo et al., 2006; Psarommatis et al., 2006; Berg et al., 2005]. In fact up to 10% of children

who fail ABR may have auditory neuropathy [Berlin et al., 2003]. Although the pathophysiology of this condition is still not fully understood, its occurrence has been associated with conditions such as hyperbilirubinaemia, ototoxic drug regimen, low birth weight, low Apgar score and anoxia. Auditory neuropathy has also been implicated in infants with no risk factors [Ngo et al 2006] as well as those with serum bilirubin levels of 16 to 25mg/dl usually not requiring exchange blood transfusion [Shapiro, 2003; Akman et al., 2004].

The high prevalence of neonatal jaundice and the associated false negative results in these babies within this population seems to draw attention to the need for AABR as a necessary component of the screening protocol especially for babies with a positive history of hyperbilirubinaemia requiring hospital admission. However, auditory neuropathy is currently not a target condition for most of the infant hearing screening programmes for well babies in developed countries and may therefore be difficult to justify in resource-poor countries. Longitudinal studies on the epidemiology of neonatal jaundice will be valuable as a necessary step for establishing its effects on learning and optimal early childhood development in Nigerian children.

5.8.3. Tracking Follow-up Compliance

The difficulties encountered with inconsistencies in the recorded names in database were the greatest challenge for tracking babies during follow-up visits. Baby's names were often different for follow up visits due to the culture of giving names only to children who survive the first week of life for the Yorubas and the first month of life for the Ibos. To resolve this problem babies were identified with their mothers' names. However, on several occasions mothers presented different names from what was held on the data base either because of change of names from maiden to married or because a family member had given a different name for the mother under emergency conditions .

The tracking efforts for defaulters showed that over half of the babies were lost to follow-up due to untraceable or fictitious contact addresses. For

instance, some addresses given by mothers were bus stops, business centres, markets and public buildings! It is surprising why such parents consented to having their babies screened when it was not mandatory to do so in the first instance. One possible reason could be due to a desire by some mothers not to be seen as being different from other mothers who consented or because they did not expect their babies to be referred since majority of babies were not.

For others, the decision to receive the first stage screening takes place at the screening site and usually does not involve the spouses. However, bearing in mind that most mothers were married, the decision to attend follow-up clinic would necessarily involve the spouses who unfortunately have not benefited from the maternal education on the importance of early hearing detection offered in the ante-natal clinics or at the screening sites. Culturally, spouses wield considerable influence on maternal health seeking behaviour especially when it affects the child. Public health campaigns would therefore need to be extended to the spouses on the benefits of infant hearing screening and this can possibly be achieved through the local electronic media.

In contrast to recorded addresses, mobile telephone numbers proved very useful in tracking defaulters. However, incomplete numbers emerged as the most common problem. Other problems encountered with tracking by mobile phone were similar to tracking with addresses and included numbers that were disconnected, business centres, family, friends, neighbours or distant relations/unknown persons. Some mothers may have deliberately given fictitious contact details as has been observed with other UNHS programmes even in developed countries [Isaacson, 2000; Finitzo, Albright & O'Neal, 1998]. Nonetheless in majority of cases, mobile phone numbers proved extremely useful in tracking follow-up appointments.

Sending a child to live with the grandmother in the village is often required as a result of maternal death or to relieve the mother from the stress of looking after a child with special needs. In this study a significant number of the

babies had been sent away to live with relations outside Lagos in the villages and towns of origin. Since it is cultural for the children to look after their ageing parents, grand parents are more than pleased to take up the role of freeing their children from further stress by helping out. Tracking follow-up defaulters further revealed that a significant number of children in this study had gone to live with their grandmothers in the villages because of maternal death or the special needs of the child.

Although in the minority, working mothers also experienced great difficulty in keeping follow-up appointments. The problems of living and working in a busy urban centre like Lagos often makes it difficult for women to keep the necessary appointments for their children without the help of friends or family. Given the diversity of factors associated with follow-up default in this study, it is unlikely that this problem can be completely eliminated. However, it would be valuable to re-emphasise the importance of follow-up to mothers whose babies failed the screening tests and verify their contact details including those of their spouses at the point of disclosing the results. Misconceptions regarding childhood hearing impairment should also be addressed as far as practicable to enhance follow-up rates [Stephens, Stephens & Eisenhart-Rothe, 2000; Odebiyi & Togonu-Bickersteth, 1987].

5.8.4. Effects of the Introduction of User Fees

As previously mentioned, all services from screening to intervention were provided free under this pilot project and there were no hidden costs of any form to parents [Khan, 2005; Nahar & Costello, 1998]. This is consistent with the practice in developed countries where infant hearing screening has been mandated as part of routine neonatal care at no cost to parents. However, an area of concern is the likely impact of introducing user fees for infant hearing screening in Nigeria in line with the current pattern of healthcare financing where patients are required to bear the (full or partial) cost of virtually all healthcare services. The introduction of user fees may become necessary for the rapid and systematic scaling up of early hearing detection and intervention services across Nigeria because of limited public funding.

However, many studies have demonstrated significant reduction in uptake of services when such user fees are introduced [James et al., 2006; Palmer et al., 2004]. The uptake of infant hearing screening is also likely to be reduced with user fees in Nigeria. For instance, a study in a private hospital in South Africa reported a drop in uptake of hearing screening from 75% when screening was subsidised to 20% when the subsidy was withdrawn [Swanepoel et al., 2007]. A similar effect is also not unexpected even in developed countries if charges for infant hearing screening services were to be introduced.

Although the lack of evidence on the extent of the likely effects of user fees may be considered as a limitation of this current study, it is necessary for Government to consider ways of mitigating their adverse effects based on the experiences with other health interventions. One option is to make all EHDI services eligible for coverage under the National Health Insurance Scheme.

In addition, in line with the stated goal of early detection of childhood hearing loss in the current National Health Policy, Government should facilitate public-private partnerships for the delivery of services especially in rural areas. The current early childhood development programmes of influential global heath actors such as the World Bank, UNICEF, WHO and UNESCO as well as the WHA Resolution on Hearing Impairment Prevention should encourage Government to actively seek financial and technical support from these multilateral institutions towards developing capacity for an effective national initiative on early detection and management of PCEHL in Nigeria.

The lack of data on the cost-effectiveness of UNHS in developing countries remains a challenge to be addressed in facilitating global investment for childhood hearing impairment within the framework of health priorities for developing countries [Cook et al,. 2006]. However, the evidence from this study and similar studies in developing countries demonstrating the exceptionally high burden of PCEHL and the need for timely intervention, particularly when this condition is being routinely detected and managed in developed countries where the burden is far less should justify immediate efforts to gather the required database on cost-effectiveness of UNHS relative to other health conditions and to other early detection options in this region.

Chapter 6

Conclusions & Recommendations

6. Conclusions and Recommendations

6.1. Research Context

Permanent childhood hearing impairment is a significant and increasing health problem worldwide with developing countries accounting for 90% of the global disease burden. Every year, between 265,000 and 798,000 babies worldwide have permanent congenital or early-onset hearing loss (PCEHL) which is associated with significant adverse impact on speech, language, cognitive and psychosocial development in early childhood. At school age, PCEHL substantially undermines educational achievement and limits vocational attainment. Affected children also suffer various forms of maltreatment such as neglect, physical, emotional and sexual abuse well into adulthood at great cost to the individual, the family and the society.

Although some of the causes of PCEHL may be prevented through improved healthcare services and practices, however, because the causes cannot be readily determined in a significant number of children, primary preventive measures are unlikely to address the full spectrum of childhood hearing impairment thereby making infant hearing screening imperative as a secondary prevention strategy. In the absence of a systematic screening for PCEHL, affected children may be detected as late as 5 years when speech and language defects become evident. In Nigeria, late enrolment in the schools for the deaf at a mean age of 10.3 years is presently the commonest mode of intervention. This late detection and intervention forecloses speech and language development for life and places the children at great risk of all forms of abuse and neglect.

Technological advances have made objective, simple-to-use, reliable and rapid tests possible for the detection of PCEHL from birth using oto-acoustic .emissions (OAE) and automated auditory brainstem response (AABR). This study has shown that these tests can be administered by non-specialists with minimal training within primary care settings to facilitate the implementation

of the resolution of the World Health Assembly (WHA) passed in 1995 urging developing countries to take appropriate steps for the early detection of PCEHL.

Although universal newborn hearing screening before hospital-discharge is now routinely offered in most developed countries, in Nigeria, like many other developing countries, majority of births occur outside hospital facilities thus making the conventional hospital-based universal newborn hearing screening inadequate or unsuitable. Consequently, a pilot project as undertaken under this research, consisting of a hospital-based and a community-based infant hearing screening programmes, was necessary to determine culturally-appropriate strategies for the early detection of PCEHL in Nigeria in line with the provisions for early identification and management of childhood hearing impairment in the country's Revised National Health Policy.

6.2. Summary of Key Findings

6.2.1. The Current Burden of PCEHL in Nigeria

The prevalence of PCEHL found in this study ranges from 5.3 to 28 per 1,000 live births which indicate that about 28,000 to 148,000 infants are born with or acquire permanent hearing loss yearly and at least 60% are of moderate-to-severe degrees. The actual rate which may be closer to the upper limit of this range is the highest ever reported in any country. This finding is consistent with various reports that have either identified Nigeria as having the highest rate of developmentally disadvantaged children in the world [Grantham-McGregor et al., 2007] or listed the country in the top league of developing nations with severe adverse perinatal conditions that portend great risk for lifelong childhood disability [Lawn, Cousens & Zupan, 2005; Black, Morris & Bryce, 2003]. This study has clearly demonstrated that PCEHL is a far more significant public health problem than previously estimated for Nigeria or when compared to countries where universal infant hearing screening is now routinely implemented.

Although, this study was conducted in Lagos, there is no evidence to suggest that the burden of PCEHL will be significantly different in other parts of the country except that higher rates are not unlikely in areas where consanguineous marriages and meningitis are common.

6.2.2. Risk Factors for PCEHL in Nigeria

It was beyond the scope of this study to establish the aetiology of PCEHL in our target population. Several known risk factors for PCEHL were identified among the 63 children detected with hearing loss out of a total of 3,333 children screened over a period of forty weeks. However, only admission in Special Care Baby Unit was strongly predictive among newborns in the hospital while birth outside hospital facilities and neonatal jaundice requiring exchange blood transfusion were predictive of PCEHL among infants attending BCG immunisation clinics. Less than 20% (14-20%) of infants with PCEHL in this study had no identifiable risk factor. The accurate and timely identification of risk factors in the remaining children remains a challenge in the absence of a universal hearing screening programme. By implication, primary prevention measures which entail overall improvement in maternal and child health care services at all levels as well as parental education should be useful but certainly not sufficient to forestall the incidence of PCEHL in this population.

6.2.3. Maternal Health-Seeking Behaviour in Nigeria

A good understanding of maternal-health seeking behaviour is crucial for the effective introduction of a new child health intervention particularly for a non-life threatening condition like PCEHL. The evidence from this study suggests that a significant proportion of mothers deliver outside hospital facilities especially in herbal homes, thus confirming the limitation of hospital-based infant hearing screening programme in Nigeria as practiced in most other countries. However, a significant proportion of mothers, regardless of where they delivered their babies, consider BCG immunisation as essential for child survival and are thus likely to visit well-baby clinics at various community

health centres where BCG immunisation is administered. Based on the results of this study, babies were predominantly more likely to be taken to the BCG immunisation clinics in their first month of life, an age when early hearing detection is highly recommended. This platform therefore offers a good opportunity to screen infants for PCEHL particularly for babies who were delivered outside hospitals and who may not otherwise be reached shortly after birth. It was also found that mothers were likely to seek both orthodox and traditional therapies for their personal health needs regardless of their educational and vocational profile.

6.2.4. Feasibility of Infant Hearing Screening in Nigeria

The conventional universal screening of newborns in hospital prior to discharge as well as the screening of infants in their first three months of life at BCG immunisation clinics at various community health centres were found to be feasible in our target population. The screening protocol consisted of a first-stage screening with (automated) transient-evoked otoacoustic emissions (TEOAE) followed by a second-stage screening with automated auditory-brainstem response (AABR) for those who were referred from the first-stage. The screening programmes were well received by parents and health workers in both settings as evidenced by the fact that no parent withheld consent to screen their baby while health workers enthusiastically assisted in educating parents on the benefits of the programme. The screening was effectively administered by non-specialist staff without prior audiological experience thus demonstrating the possibility of offering this service widely at the primary care level. Targeted screening based on relevant risk factors in the hospital or the community was not prospectively evaluated. However, a retrospective analysis of our results would suggest that screening newborns admitted to Special Care Baby Unit may be considered as an interim strategy for a hospital-based screening programme with the likelihood of detecting about 40% of babies with PCEHL. Similarly, offering hearing screening to infants who were born outside hospitals or those with a history of neonatal jaundice requiring exchange blood transfusion may "theoretically" lead to the detection of at least two-thirds of

infants with PCEHL. However, these outcomes would need to be validated in future studies bearing in mind the practical difficulties that may be encountered in accurately identifying eligible infants at the various community health centres and the stigma often associated with childhood deafness.

6.2.5. Performance of Screening Options & Cost Implications

A screening coverage (i.e. percentage of eligible babies actually screened) of 97% was achieved for the hospital-based programme and 100% for the community-based programme, both of which exceeded the common international benchmark of 95% [JCIH, 2000]. The initial referral rates of 3.5% and 4.4% for the hospital-based and community-based programmes respectively also fell within acceptable range of the 4.0% target recommended by experts [JCIH, 2000].

However, the return rates for follow-up tests after the initial TEOAE test were generally poor, particularly among mothers in the hospital-based programme for reasons that warrant further investigation and appropriate action. About 16% of mothers whose babies were referred for diagnostic test in the hospital-based programme returned compared with 61% in the community-based programme. The international benchmark of 70% [JCIH, 2000] was not achieved in either programme. It was very much unlikely that the cost of transportation was a contributory factor as free transportation was provided from the sites for the initial screening to the referral/diagnostic centre. Similarly, the average age of confirmation of hearing loss was 8 months for babies from the hospital-based programme compared with 2 months for infants under the community-based programme. If intervention were to be initiated by 6 months as recommended in developed countries, babies in the hospital-based programme would have been enrolled late. The yield for PCEHL under the community-based programme of 22.5 per 1,000 is more-than four-fold of the yield of 5.3 per 1,000 from the hospital-based programme.

A comprehensive cost-effectiveness of the screening options was outside the scope of this study. However, a comparative rudimentary analysis of the cost per screening a baby showed that the community-based programme (US$7.62 or £3.91) was about half the cost in the hospital-base programme (US$13.30 or £6.82). Targeted screening emerged as a more expensive option, costing about US$85.91 or £44.05 to screen a high-risk baby in the hospital based-programme compared with US$27 or £13.85 to screen a high-risk infant in the community-based programme.

6.2.6. Challenges for Infant Hearing Screening in Nigeria

The first challenge related to the high ambient noise levels at our screening locations, which are characteristic of urban centres. Unfortunately, most of the current objective screening technologies for detecting PCEHL were designed for environments with significantly lower ambient noise levels. Consequently, the performance and suitability of various models of otoacoustic emissions was contingent upon their in-built tolerance limits within a range of ambient noise levels. Finding a suitable site or section within a screening location either in the hospital or in a community health centre was necessary but sometimes quite challenging especially where there is acute shortage of space.

Another challenge was how to identify children with auditory neuropathy who are usually missed by any protocol that is based on TEOAE only or requiring a first-stage TEOAE screening before further evaluation (with AABR and/or ABR). This is a limitation that cannot be readily overcome in practice in any country and therefore may not merit a serious consideration at this stage.

Effective tracking of parents of infants who require follow-up screening or evaluation was a challenge in this study as with programmes even in developed countries. Efforts to minimise this problem would require on-going education of mothers and their spouses supported with dedicated full-time staff. The complete elimination of this problem is perhaps unattainable

especially in a developing country like Nigeria to a variety of logistical constraints.

Lastly, the excellent coverage achieved for the initial screening was not unconnected with the fact that parents were not charged for the services. It is not unlikely that screening uptake will be reduced where the introduction of user fees are necessary to sustain the availability of these services. There is therefore a need to explore ways of mitigating this threat as far as practicable.

6.3. Conclusions

Universal newborn hearing screening has emerged as an essential component of neonatal care in developed countries which only account for about 10% of the global burden of PCEHL. Current evidence indicates that infant hearing screening programmes are also progressively being undertaken in a growing number of developing countries in recognition that PCEHL is a significant public health condition. The timely implementation of these recommendations in Nigeria, a country with one of the highest rates for PCEHL and that presently accounts for the highest proportion of developmentally disadvantaged children globally, is not only vital for the optimal management of permanent childhood hearing impairment as envisioned by the Revised National Health Policy for Nigeria but is also consistent with the early childhood development priorities of the World Bank, UNICEF, WHO and UNESCO as well as the attainment of the first two millennium development goals on poverty reduction and universal primary education for all. The choice of an appropriate infant hearing screening model based on all key operational efficiency factors including the cost per screening a child weighs heavily in favour of community-based programme. So also is the consideration for high rate of non-hospital deliveries and the associated adverse postnatal conditions which cannot be significantly altered or curtailed readily in Nigeria. Finally, the high prevalence of infants with a history of neonatal jaundice often requiring exchange blood transfusion as

found in this study, which can be effectively curtailed, merits urgent attention particularly in view of a lack of global priority for this condition presently.

6.4. Recommendations

1. Infant hearing screening should be considered as an essential public health programme to be incorporated into the current Expanded Programme on Immunisation for Nigeria in line with the provision for early hearing detection in the Revised National Health Policy.

2. The Federal Government should commission additional pilot studies in the five remaining geo-political regions of the country and among the rural populations to establish more accurately the implications of the findings in this study throughout Nigeria. The outcomes of these pilot studies should provide a basis for a comprehensive national assessment of the required improvements in intervention services (including educational support) for children with hearing impairment.

3. Further studies would also be useful to ascertain if targeted newborn hearing screening of SCBU babies, in which screening is entrusted to staff nurses as part of their routine duty, is feasible and cost-effective as an interim strategy where UNHS is not immediately practicable.

4. There is need to establish other possible risk factors among newborns at birth through a wider hospital-based multi-centre research while further community-based studies are required to establish more accurately the prevalence, aetiology, pattern and management of neonatal jaundice in Nigeria as well as the causal pathway for PCEHL associated with births without skilled attendants to guide appropriate primary prevention initiatives.

5. There is an urgent need for government at all levels to invest in capacity-building for audiological services in public hospitals and community health centres as well as to actively promote parental

education on the significance of early hearing detection and intervention at the community level regardless of where babies are born to facilitate prompt diagnosis of PCEHL and minimise follow-up default rates. Partnerships with non-governmental organisations for the development and provision of early hearing detection and intervention services as demonstrated in this pilot project should also be encouraged and supported.

6. While the cost of screening an infant is significantly lower than the rates in developed countries, further studies are valuable to determine the possible effects of the introduction of user fees at the population level. Moreover, beyond the cost of screening a baby derived from the limited cost analysis in this study, it will be useful to undertake a more robust cost-effectiveness analysis of the hospital-based and community-based screening programmes to guide possible future public sector financial investment in early hearing detection and intervention services, and the systematic scaling-up of this pilot programme throughout Nigeria possibly within the stepwise framework recommended by WHO [Epping-Jordan et al., 2005]. Moreover, the Federal Government should urgently consider making early hearing detection and intervention services eligible under the National Health Insurance Scheme to mitigate the possible effects of the introduction of user fees where these services cannot be provided or sustained otherwise.

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Appendices

Appendix 3.1

Great Ormond Street Hospital for Children NHS Trust / The Institute of Child Health Local Research Ethics Committee

Institute of Child Health
30 Guilford Street
London
WC1N 1EH

Tel: 020 7905 2620
Fax: 020 7905 2201
Email: l.howe@ich.ucl.ac.uk

9th January 2004

Dr B Olusanya
Audiological Medicine
ICH

Dear Dr Olusanya,

Title: **A national programme for newborn hearing screening in Nigeria**
R&D registration number: **03AM04**
Protocol number/version: **N/A**

Letter of Support for Clinical Research undertaken in Other Countries

The above research has been reviewed by the Chairman of the Great Ormond Street Hospital for Children NHS Trust / Institute of Child Health Research Ethics Committee. The Chairman has agreed to issue a letter of support in line with the Committee's procedures for the appraisal of clinical research projects based in other countries. The scope of this letter is described more fully below:

1. Ethical 'support' is given to studies carried out overseas involving GOS/ICH staff or premises, which the GOS/ICH REC believe comply with broadly stated ethical standards. The GOS/ICH REC do not feel that it is within their remit, nor do they have the training or skills, to assess the full ethical impact of studies carried out overseas. However, the Chairman has considered whether the scientific methods are sound and suitable for the aims of the research, whether drugs or vaccines or devices to be studied meet adequate standards of safety (as applicable), whether there is sound justification for conducting the research in the host country, and consider that the proposed research does not in principle violate the ethical standards applied to UK projects. A letter of 'support' from this Committee is evidence that the research proposal has been considered in that ethical framework and found to be acceptable, but must be regarded as lacking the full authority implied by 'full approvals' given to UK projects in similar circumstances.
2. The full ethical impact of the study must be considered and approved by an appropriate body in the host country. This body is relied upon to have special responsibility to determine whether the aims of the research are responsible to the needs and priorities of the host country, for assuring that procedures for selection of subjects, the plans for obtaining informed consent and for ensuring privacy and confidentiality and the level of inducement offered are ethically acceptable.
3. Ethical support is given for a period of xxxx months from the commencement of the project. If you wish to start the research more than twelve months from the date of this letter or extend the duration of your 'support' you should seek Chairman's approval.
4. You must seek Chairman's support for proposed amendments to the research for which this approval has been given. Ethical support is specific to this project and must not be treated as applicable to research of a similar nature, e.g. using the same procedure(s) or medicinal product(s). Each research project is reviewed separately and if there are significant changes to the research protocol, for example in response to a grant giving body's requirements, you should seek confirmation of continued ethical support.
5. It is your responsibility to notify the Chairman immediately of any information which would raise questions about the safety and continued conduct of the research.

Yours sincerely

Laura Howe
Research Ethics Coordinator
cc. Prof L Luxon

Appendix 3.2

LAGOS STATE GOVERNMENT

P. M. B. No.
Telephone
Telegrams
All Letters to be addressed to the Permanent Secretary
Our Ref. No. SHMB/728T

Hospitals Management Board
26, Catholic Mission Street,
Lagos.

Date: 17th December, 2003.

Dr. Bola Olusanya,
Consultant Paediatrician (Audiological Medicine)
C/o Institute of Child Health
And Great Ormond Street Hospital for
Children NHS Trust

RE: ETHICAL CLEARANCE TO USE STATE HOSPITALS

I am hereby directed to convey to you the approval of the Permanent Secretary to carry out your research on the desired topic in the following hospitals:

1. Lagos Island Maternity Hospital, General Hospital Ebagada, Massey Street Children's Hospital, General Hospital, Surulere, Apapa Health Centre, Orile Agege General Hospital, Harvey Road Hospital, Onikan Health Centre, General Hospital Ikorodu, General Hospital Epe and General Hospital Badagry.

Please liase with the Medical Directors of the hospital when you get there.

A Amaylge 17/12/03
Dr.(Mrs) A.M. Omeyige
for: Permanent Secretary

Appendix 3.3

Training Programme for Screening Staff

Curriculum and Guidelines

Theoretical Session

1. Normal basic anatomy and physiology of the ear, speech and language development.
2. Checklist of indicators of normal hearing and the at-risk signs of possible hearing impairment.
3. Early warning signs of hearing impairment.
4. Consequences of hearing loss and the benefits of early detection and appropriate intervention.
5. Knowledge of the benefits of objective screening methods over the subjective tests.
6. The relationship between the various units involved with newborn screening and the importance of coordinated hospital- and community-based programmes.
7. Appreciate that the efficiency of the screening programmes are dependent on the ability to accurately track all births and screen up to 95%.

Practical sessions for skill acquisition

1. Competent use of the test protocol and the appropriate and accurate use of the checklist.
2. Ability to carry out the screening tests accurately with minimal discomfort to the baby
3. Accurate interpretation of test results and knowing when a retest is necessary without increasing parental anxiety unduly.
4. Understanding that the ability to institute appropriate action depends on accurate interpretation of the results of the hearing-screening test.
5. Develop their ability to manage the data collected through accurate record keeping.

6. Develop their ability to convince those referred for follow-up tests of the importance of keeping their appointments.

Tips for proper probe fit

Proper probe fit is the key to obtaining a good OAE screening test. A tight seal ensures that your screening test will be quick and accurate. Some tips for obtaining a good probe fit are:

- Select the largest probe tip possible. A probe tip that is too small will allow too much environmental noise in.
- Gently tug on the probe once it is in the ear to make sure there is resistance. If it slides out easily, it is too loose.
- Newborns often have collapsed ear canals. This can especially be problematic if the baby has just been lying on the ear you wish to test. To fully open the ear canal and obtain a good probe fit:
    - Gently massage the area in front of the ear in a circular motion for 10-20 seconds before inserting the probe tip.
    - Gently pull up and back on the pinna, or outer ear, and massage/rotate gently a few times before inserting the probe.
    - When inserting the probe into the ear, pull up and out gently on the pinna, or outer ear to fully open the ear canal.
- Having the baby swaddled during testing will help to make sure that it doesn't knock the probe out of its ear during the test.
- Don't hold the probe during testing. This can cause the probe to touch the wall of the ear canal and prevent a signal from getting through. It can also cause noise that interferes with testing.
- Make sure your probe tip is clean — sometimes, the probe tip may become clogged by vernix or debris. If you need to clean the probe tip, be sure to remove it from the probe prior to cleaning, so you don't inadvertently push the vernix or debris into the probe.

Guidelines for optimal test environment

- Quiet room
- Quiet, sleeping baby
- Ideally, conduct the test after the baby has been fed
- Swaddled baby
- Clean diaper
- Post a sign to inform others that a hearing test is in progress
- Complete the test prior to discharge
- In conjunction with nursing staff, establish test times that are optimal for nurses, parents, and babies' schedules.

Guidelines for Troubleshooting with Equipment

TEOAE

1. Have you chosen the right size probe tip?

Ideally, the largest probe tip that fits into a baby's ear should be used in order to create a good seal. This helps to keep out extraneous environmental noise. A probe tip that is too small will allow too much noise in, which prevents the emissions from being recorded.

2. Is the probe tip clogged?

Often, babies have vernix or debris in the ear canal. This can clog the probe tip, which may affect the stimulus going into the ear. Ensure that the probe tip is clear of debris from the baby's ear canal. Also, check the probe tip to see if you can hear the stimulus.

3. Is the probe tip all the way in the ear?

Often, a baby's ear canal may be collapsed. This can especially be a problem if the baby has just been lying on the ear you are trying to test. To help ensure a good probe fit, gently massage the area in front of the baby's ear in a circular motion for 10 seconds prior to inserting the probe tip. Also, try pulling up and back on the pinna, or outer ear, and massage/rotate gently a few times before inserting the probe. When inserting the probe into the baby's ear, gently pull up and back on the pinna or outer ear to fully open the

ear canal and help obtain a good probe fit. A good self-check is to gently tug on the probe once it is in the ear. There should be resistance; if it slides out easily it is too loose.

4. Are you consistently getting more refer results for the second ear?

Many times the second ear can be more difficult to test because the baby has just been lying on it. Because a newborn's ear canals are soft and sticky, this can easily cause the baby's ear canal to collapse. If you are still having problems screening the second ear after following the suggestions in the above paragraph, try leaving the baby with that ear up, or lying on his or back for a while and come back later and try again. This will give that ear a chance to open up and dry out.

5. Is the baby resting quietly?

Swaddle the baby to keep him/her from knocking the probe from the ear. An optimal time for testing is just after a baby is fed, and when they have a clean diaper. Excessive movement, sucking, or crying will interfere with the test.

6. Have you minimized other room noise?

Whenever possible, eliminate extra noise from monitors or other equipment. Check with the nursing staff before touching any medical equipment.

7. Check for a proper stimulus level.

Regular equipment checks and annual calibration helps to ensure that equipment remains in good working order.

8. Don't hold the probe while testing

You shouldn't need to hold the probe while testing if you have a good fit. Holding the probe during testing may cause it to touch the wall of the ear canal and prevent the signal from getting through. You can also cause noise that interferes with the testing.

AABR

Check impedance levels.

If the levels are unacceptably high, re-scrub the baby's skin and make sure that the electrodes are plugged in.

1. Is the baby sleeping quietly?

If the baby is restless or crying, you will not get a good test. The electrodes pick up small impulses from the hearing nerve, but they also pick up muscle movements (artifacts). The baby should be swaddled and sleeping/resting quietly for a good test.

2. Are there other pieces of medical equipment nearby that may cause electrical interference?

Whenever possible, eliminate extra noise from monitors or other equipment. Check with the nursing staff before touching any medical equipment.

Communicating results to parents

It is important that information about the hearing screening and the results are conveyed to the parents in a professional, thoughtful and sensitive manner. By saying, *"Your baby didn't pass the hearing screen"* you may have just changed this family's whole life.

1. Points to remember when communicating results to parents:

Information should be given both verbally and in writing. Give every parent the Early Help Better Future screening leaflet. Remind them to not to leave the leaflet in the hospital or screening site.

Inappropriate communication of screening results can cause undue stress and anxiety for the parents. Remember to use the proper terminology. Never say *"fail"*. If a baby does not pass the hearing screening, the term *"refer"* should be used instead.

This is a emotionally sensitive time for parents — information should be conveyed in a supportive, confidential environment. Make sure information is conveyed in an unhurried manner, with plenty of time allowed to answer questions

2. If a baby is referred for further testing

If a baby does not pass the hearing screening, it is crucial NOT to use the words *"failed"* or *"did not pass"*. This terminology implies to the parents that their baby has a hearing loss, or is Deaf.

Instead, you should say, *"We are referring your baby to a nearby Specialist Hearing Centre for further testing,"* or *"We are referring your baby for a re-screening because the test results were inconclusive today."*

Parents must be made aware that newborn hearing screening is designed to catch babies who are at risk for hearing loss and need further testing. Remind parents that hearing loss does not necessarily equal deafness.

Hearing loss can range in severity from mild to severe-profound (deaf). Further diagnostic testing is needed to confirm their baby's hearing status.

If a baby is referred, the family should be informed that there could be several reasons why their baby is being referred for further testing.

The most common reasons are:

- An ear canal blocked with debris (most common)
- The presence of middle ear fluid
- A permanent hearing loss (approximately 3 in 1000 births)

When screeners discuss results with families they should be careful not to downplay a refer result, while at the same time being careful not to panic the family.

IF IN DOUBT, CONTACT THE COORDINATOR!

Training Programme: Main Themes and Methods

KNOWLEDGE	SKILL	ATTITUDE	METHODS
The programme is designed to work within the hospital environment and not in isolation.	Ability to work together with other health workers for the success of the programme.	Understand the need to successfully implement this programme within the hospital framework.	-Lectures -Video -Role play
Normal Hearing and Development of Speech and Language Skill.	Ability to fill the questionnaire form and use the checklist accurately and appropriately.	Appreciate the need for a family-friendly programme in the overall success of the study.	- Lectures - Demonstrations - Group work - Assignments
-Check list of indicators of normal hearing. -The at-risk signs. -The early warning signs of hearing loss. - Advantages of using objective tests over the subjective ones.	Ability to use test- protocol and conduct TEOAE and AABR tests accurately with minimal discomfort.	Understand the relative importance of the prevailing cultural and religious issues.	-Video demos -Role play -Practical demo -Practical Sessions -Assessments
Consequences of hearing loss and the benefits of early detection and intervention initiatives.	Ability to educate parents on the consequences of hearing loss and the value of NHSP without unduly increasing their anxiety.	Understand that the NHSP will be associated with some degree of parental anxiety and stress.	-Clinical demo -Role play -Practical Sessions -Assessments
The importance of tracking referrals to the overall success of the programme.	Ability to manage data and ensure that referrals keep their appointments	Embrace the challenge of follow-up and tracking of all referrals.	-Clinical demo -Practical Sessions -Assessments

Appendix 3.4

Early Help Better Future Programme

Speech and Language Milestones for your Baby

Birth to 3 months

- Recognises and quiets to parent's voice
- Startles to loud sounds
- Laughs, gurgles and coos

3 to 6 months

- Awakens to sounds or speech
- Turns towards interesting sounds
- Makes a variety of sounds enjoys babbling

6 to 12 months

- Understands first words such as "Da-Da", and "stop it", "Go", "Come"
- Responds to his or her name
- Enjoys sounds from rattles, and similar toys
- Coos to music and imitates speech

12 to 18 months

- Says first words such as "Da-Da", "Ma-Ma", "Bye-Bye"
- Identifies body parts and favorite toys by pointing to them
- Responds to sounds coming from far away in all directions

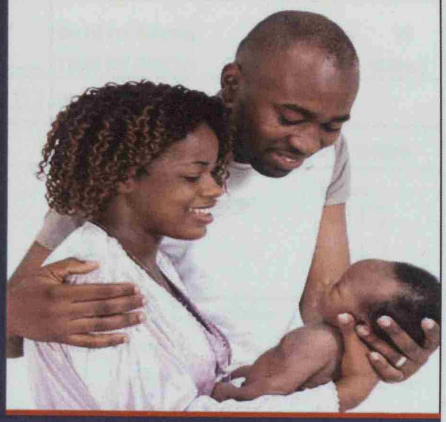
18 to 24 months

- Has a vocabulary of few words
- Speaks two word phrases
- Understands simple "yes" and "no" questions
- Refers to self by name
- Follows simple directions

24 to 3 years

- Has vocabulary of many words by 3 Years
- Speaks to communicate needs, wants and experiences
- Speaks simple sentences
- Recognises different sounds
- Understands most of what is said to him or her

INFANT HEARING SCREENING IN NIGERIA



A Sound Start for Every Child

Early Help Better Future Programme

Congratulations on the birth of your baby!!!

A baby may be born with a hearing defect due to nobody's fault. This defect may not be noticed for months until perhaps the parent observes that the child does not react to a loud sound or noise. A normal hearing child would progressively react to sound from birth

A child with a hearing defect will not develop age-appropriate speech and language if not detected and helped early. This is why this newborn hearing screening has been introduced to assure parents before or shortly after leaving the hospital that their baby was not born with a hearing defect that may affect their future well-being. It is one of the essential routine checks in the first few days of life. It has now become the standard practice in maternal and child care in advanced countries.

Will the test hurt my baby?

No. The procedure is quick and painless and can be done while your baby is asleep. No injection, drugs or sedatives will be given to your baby for this test. You may in fact observe the screening test while it is being done.

When will the screening test be done?

A medical staff will contact you to arrange for a suitable time to screen your baby as soon as possible. Every effort will be made to ensure that your baby is tested before you leave hospital or shortly thereafter.

What happens after the screening test?

The medical staff that screened your baby will tell you the result immediately. You will be notified if your baby requires additional test. It is important that you keep any appointment given to you by the medical staff to have your baby properly tested and supported in every way possible.

Infant Hearing Screening Programme

Parental CONSENT Form

Child's Name: _____

Hospital: _____

Date and Time of Birth: _____

Parents Surname: _____

Mother's Name: _____

Declaration

A medical staff has explained to me the importance of a hearing screening test for my baby.

I would like to have my baby's hearing tested. I would also like to have an appointment to see a medical staff should additional tests be required.

Signed: _____ Date: _____

Medical Staff: _____ Date: _____

Baby ID. Number: _____

Please complete this page, detach and give to the medical staff before screening

Appendix 3.5

INFANT HEARING SCREENING PILOT PROJECT, NIGERIA

MEDICAL RECORD I

Tick (✓) the right box and fill in the gaps

FORM A MATERNAL RECORD

NURSERY TYPE: WBN ☐ NICU ☐ PAED WARD ☐ OPD ☐ CHER ☐

IDENTIFICATION NUMBER: _____

PLACE WHERE BABY IS BORN:
Govt Hospt. ☐ Health Centre ☐ Private Hospt. ☐ Home ☐ Herbal Home ☐ Church ☐ Other ☐

ETHNICITY: YORUBA ☐ HAUSA ☐ IBO ☐
MINORITY ☐ OTHERS ☐

CONTACT'S NAME LAST: FIRST: **RELATIONSHIP TO BABY** _____

DATE OF BIRTH: DD MM YY
TIME OF BIRTH: HR MN (24hrs.)

CONSANGUITY - Both Parents are: Unrelated ☐ Cousins ☐ Uncle/Nice ☐ Distant Relatives ☐

INBORN: YES ☐ NO ☐

BABY'S SEX: FEMALE ☐ MALE ☐ UNDETERMINED ☐

GESTATIONAL AGE: _____ WEEKS:

MOTHER'S AGE: _____ YRS
AGE AT MARRIAGE: _____ YRS
GRAVIDA _____
PARITY _____

ATTENDANT AT DELIVERY:
None ☐ Doctor ☐ Midwife ☐ Trained Nurse ☐
Auxiliary Nurse ☐ TBA ☐ Neighbour ☐
Relative/Family Member ☐ Specify _____

TWINS/Multiple:
YES ☐ If Yes: 1st ☐
NO ☐ 2nd: ☐
Others ☐

THE BABY'S CORD WAS CUT WITH: Old Blade ☐ New Blade ☐ Clean Scissors ☐ Sterile Scissors ☐ Other:
UMBILICAL CORD WAS DRESSED WITH: Nothing ☐ Spirit ☐ Mentholated Powder ☐ Other:
BABY WAS DRYED AND WRAPPED: Immediately ☐ Within 1 hour ☐ After 1 hour ☐ Specify: _____
BABY'S FIRST BATH : Immediate ☐ Within 1 hour ☐ Within 6 hours ☐ Over 12 hours ☐
METHOD OF FEEDING: Exclusive Breast Feeding ☐ Bottle Feeding ☐ Breast milk & Bottle ☐

RELIGION:	None	Christianity	Islam	Traditional	Other
FATHER	☐	☐	☐	☐	☐
MOTHER	☐	☐	☐	☐	☐

MATERNAL HIV STATUS
- VE ☐
+VE ☐
UNKNOWN ☐

EDUCATION:	None	Primary	Secondary	College	University
FATHER	☐	☐	☐	☐	☐
MOTHER	☐	☐	☐	☐	☐

HOSPITAL STAY > 5 DAYS
NO ☐
YES ☐ specify

OCCUPATION:	None	Small Trade	Pepper Grinding	Daily Paid	Regular	Management
FATHER	☐	☐	☐	☐	☐	☐
MOTHER	☐	☐	☐	☐	☐	☐

MOTHER IS:
MARRIED ☐ DIVORCED ☐
UNMARRIED ☐ WIDOWED ☐

HOUSING:	Temporary	Brick	Shared Sanitation	Owner	Rented
	☐	☐	☐	☐	☐

MATERNAL HEIGHT: CM
Tall ☐ Average ☐ Short ☐

POSSESSIONS:	None	Radio	Television	Mobile Phone	Land Phone	Car
	☐	☐	☐	☐	☐	☐

MATERNAL COMPLICATIONS:
VVF ☐ PPH ☐ DEATH ☐

PRIOR TO THIS PREGNANCY DID BABY'S MOTHER HAVE ANY OF THE FOLLOWING?

RH INCOMPATIBILITY ☐	HYPERTENSION ☐	DIABETES ☐	PREVIOUS STILL BIRTH ☐
HERPES SCREEN ☐	RUBELLA IMMUNIZATION ☐	SYPYLIS SCREEN ☐	SICKLE CELL DISEASE ☐

WHILE PREGNANT DID BABY'S MOTHER HAVE ANY OF THE FOLLOWING?

ANTE-NATAL CARE ☐	CHICKEN POX ☐	MEASLES/GERMAN MEASLES ☐	JAUNDICE ☐	MALARIA ☐
DIABETES ☐	BLEEDING ☐	PRE-ECLAMPSIA ☐	DRUG ABUSE ☐	ECLAMPSIA ☐
PREGNANCY INDUCED HYPERTENSION ☐	SYPHILIS ☐	MEDICATION ☐	ANAEMIA ☐	RASH IN PREG ☐

HAVE ANY OF BABY'S RELATIVES HAD PERMANENT HEARING LOSS AS A CHILD (<7 YEARS)

Mum	Dad	Mum's Mother	Mum's Father	Dad's Mother	Dad's Father	Sister	Brother	Other	Specify	Not Sure	No
☐	☐	☐	☐	☐	☐	☐	☐	☐		☐	☐

Age of Last Child _____

AGE WHEN HEARING LOSS STARTED ☐ ANY EAR SURGERY ☐ RELATIVE HAS OR HAD HEARING AID ☐ RELATIVE HEARS WELL NOW ☐

RISK FACTORS: +ve Family History ☐ Maternal Rash in Pregnancy ☐ Maternal Illness in Pregnancy ☐

COMPLETED BY: **SIGNATURE:** **DATE:**

INFANT HEARING SCREENING PILOT PROJECT, NIGERIA

MEDICAL RECORD II

Tick (✓) the right box and fill in the gaps

FORM B INFANT RECORD

PLACE WHERE BABY IS BORN:

NURSERY TYPE: WBN ☐ NICU ☐ PAED WARD ☐ OPD ☐ CHER ☐

IDENTIFICATION NUMBER: _____

DATE OF BIRTH: DD MM YY

BABY'S NAME LAST: FIRST:

TIME OF BIRTH: HR MN (24hrs.)

BABY'S SEX: FEMALE ☐ MALE ☐ UNDETERMINED ☐

INBORN: YES ☐ NO ☐

BIRTH WT _____ GM | **LENGTH** _____ CM | **HEAD CIRCUM** _____ CM | **DATE OF EXAM** ____DD ____MM ____YY

GESTATIONAL AGE: _____ **WEEKS:**

GENERAL EXAMINATION IS SATISFACTORY: YES ☐ NO ☐

MATERNAL HIV STATUS - VE ☐ +VE ☐ UNKNOWN ☐

HOSPITAL STAY > 5 DAYS NO ☐ YES ☐ specify:

TWINS/Multiple: YES ☐ NO ☐ 1st: ☐ 2nd: ☐ Others ☐

MODE OF DELIVERY: SVD ☐ ELECTIVE C/S ☐ EMERGENCY C/S ☐ FORCEPS ☐ VACUUM ☐ ASST BREECH ☐

CONTACT:

Mother ☐ Father ☐ Grandmother ☐

Relation ☐ Specify _____

ANY SUSPECTED SYNDROME NO ☐ YES ☐ Specify:

PRESENTATION: CEPHALIC ☐ BREECH ☐ FACE ☐ OTHERS ☐ Specify:

CONTACT ADDRESS:

APGAR SCORE: ONE MINUTE _____ FIVE MINUTES _____

ACTIVE RESUSCITATION OVER 5 MINUTES NO ☐ YES ☐ Specify Medication _____

GSM: _____

TEL: _____

HEARING TEST

	RIGHT	LEFT
OAE 1	☐	☐
OAE 2	☐	☐
AABR	☐	☐

DEFECTS OF HEAD & NEC NONE ☐ CEPHALOHAEMATOMA ☐ DYSMORPHIC ☐ CLEFT LIP/ PALATE ☐

EAR CANAL	LOW SET EARS	PITS	SKIN TAGS
Right ☐	Right ☐	Right ☐	Right ☐
Left ☐	Left ☐	Left ☐	Left ☐

OTHER ANOMALIES CARDIAC ☐ UNDESCENDED TESTIS ☐ RENAL ☐ SPINA BIFIDA ☐ HYDROCEPHALUS ☐

DEFECT Up-LIMB ☐ DEFECT Lo-LIMB ☐ CLUB FOOT ☐ OTHERS ☐ Specify: _____

PERI-NATAL CONDITIONS: **Awareness of NHS** ☐ During ANC ☐ Other ☐ Specify: _____

PROLONGED LABOUR ☐	CYVICAL DYSTOCIA ☐	ASPHXIA/HYPOXIA ☐	ANAEMIA ☐
PROLONGED RUPTURE OF MEMBRANES ☐	CORD PROLAPSE ☐	HYPOGLYCAEMIA ☐	NEONATAL TETANUS ☐
CEPALOPELVIC DISPROPORTION ☐	CORD ROUND THE NECK ☐	CONGENITAL MALARIA ☐	ANTEPARTUM HAEMORRHAGE ☐
PLACENTA PREVIA ☐	MECONIUM LIQOUR ☐	SEIZURES ☐	PREVIOUS C/S ☐
OBSTRUCTED LABOUR ☐	MECONIUM ASPIRATION ☐	MENINGITIS ☐	TRIAL OF LABOUR ☐
FOETAL DISTRESS ☐	RESPIRATORY DISTRESS SYND ☐	NEONATAL SEPSIS ☐	OXYTOCIN INDUCED ☐

OTOTOXIC MEDICATION: NONE ☐

AMINOGLYCOSIDE _____ LOOP DIURETICS _____

DURATION _____ DURATION _____

HERBAL DRUGS IN PREGNANCY: Occasionally ☐ Frequently ☐ Not at All ☐

ALCOHOL INTAKE IN PREG: Occasionally ☐ Frequently ☐ Not at All ☐

SMOKING IN PREGNANACY: Occasionally ☐ Frequently ☐ Not at All ☐

NEONATAL JAUNDICE

- HYPERBILIRUBINAEMIA: ☐
- EXCHANGE TRANSFUSION: ☐
- PHOTOTHERAPY: ☐

HIGHEST UNCONJUGATED LEVEL _____ mg/dl

OTHERS _____ specify

Intrapartum Maternal Fever ☐

Malaria Diagnosis +ve ☐

RISK FACTORS: DEFECTS OF HEAD AND NECK ☐ PREMATURITY/SMALL FOR GESTATIONAL AGE ☐ HOSPITAL STAY >5 DAYS ☐ NEONATAL JAUNDICE ☐

COMPLETED BY: **SIGNATURE:** **DATE:**

NEXT APPOINTMENT: DD _____ MM _____ YY _____

Appendix 4.1

Notes to Comparative Cost Analysis of Hospital- and Community-based Screening Programmes

Unit cost of Echo-Screen TS [plus Shipping &Duty to Lagos] - $3,750

- Unit cost of Algo Portable [plus Shipping & Duty to Lagos] - $15,625
- Computer plus Printer, Accessories & Software - $2,000

The total cost of 3 units of Echo-Screen, 2 units of Algo Portable and 1 Computer System for Data Management [$44,500] was shared equally between the two sites. The third Echo-Screen was used as back-up unit for both sites. All equipments were amortized over 5 years or 260 weeks and pro-rated for the duration of screening. However, Lin et al [2005] have reported average life-span of 5 to 7 years for screening equipments in a comparable programme that started since 1998.

Note 2:

The research staff were remunerated at following rate in local equivalent:

- One Coordinator - $9,000 per annum
- Two screeners - $3,000 per annum each.
- Two support staff - $2,400 per annum each
- One Back-up staff - $2,400 per annum
- Part-time Data Entry Clerk - $1,500 per annum

The total staff expenditure of $18,230 was split equally for both sites.

Note 3:

Stationery expenditure included the cost of printing the questionnaire/data collection card, information brochure on the screening programme with a detachable consent form and display posters. The total unit cost was computed as $0.15 per person.

Note 4:

The ear tips for the Echo-Screen cost about $160 for a pack of 250 pieces. Because they were reusable, the cost per baby was worked out at around $0.10.

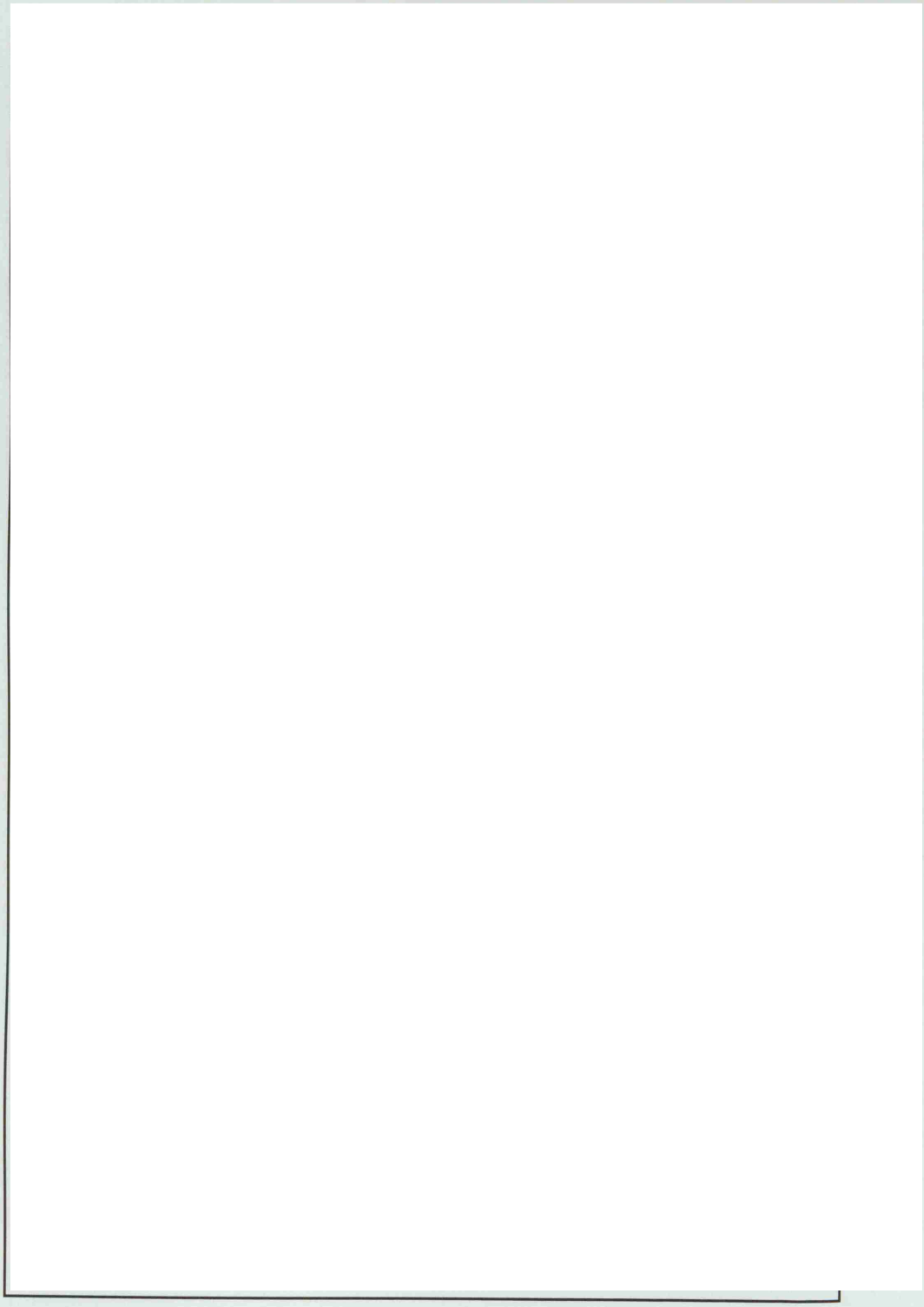
Note 5:

The consumables for AABR consisted of a set of disposable earphones and sensors/electrodes which was computed as $12 per baby, including shipping and duty charges to Lagos. Please note that unlike TEOAE which was done for all babies, this cost was only incurred for babies who failed TEOAE test.

Note 6:

The transportation cost for mothers of infants referred at community screening sites for AABR was estimated as $4.00 per person to cover the running cost of transporting the parents to the audiological centre where the test was done.

Appendix 5.1



A traditional method for settling a restless baby

Appendix 5.2

Scenario Analysis for Community-based Screening

Measures	If the initial Central Screening was never introduced	If 100% (not 75%) of Eligible Infants were screened
Number of babies screened		
• TEOAE	2,277	3,036
• AABR [6.9%]	158	210
Fixed Costs	US$	US$
• Equipment [note1]	3,423	3,423
• Staff Remuneration [note 2]	9,115	9,115
Sub-Total	12,538	12,538
Variable Costs		
• Stationery [note 3]	342	455
• TEOAE Tips [note 4]	228	304
• AABR Electrodes, etc [note 5]	1,896	2,520
• Transportation [note 6]	632	840
Sub-Total	3,098	4,119
Total Cost	15,636	16,657
Screening Cost per Baby	US$6.87 [£3.52]	US$5.49 [£2.82]

Please note:

- This computation assumes no changes in the composition of the current screening team as in Appendix 4.1.
- The average no of babies screened per day was 13 for the 2003 infants covered in the study, based on the 4 days of screening per week. At the current level of fixed cost it is possible for the same team to expand coverage to 3036 infants at an average of 13 babies per day, by increasing screening from 4 to 6 days per week.
- See Appendix 4.1 for description of Notes 1- 6.

Appendix 5.3

Scenario Analysis for Targeted Screening based on Predictors of PCEHL

Measures	Hospital-based	Community-based
Number of babies screened		
• TEOAE	180	1,166
• AABR [100%]	180	1,166
Fixed Costs	US$	US$
• Equipment [note1]	3,423	3,423
• Staff Remuneration [note 2]	9,115	9,115
Sub-Total	12,538	12,538
Variable Costs		
• Stationery [note 3]	27	175
• TEOAE Tips [note 4]	18	116
• AABR Electrodes, etc [note 5]	2,160	13,992
• Transportation [note 6]	720	4,664
Sub-Total	2,925	18,947
Total Cost	15,463	31,485
Screening Cost per Baby	US$85.91 [£44.05]	US$27.0 [£13.85]

- See Appendix 4.1 for description of Notes 1- 6.

Appendix 5.4

Counselling session with a group of parents of infants confirmed with hearing loss

Related Research Work: [January 2003 – April 2007]

Publications in Peer-reviewed Journals

1. Olusanya BO. (2007). **Promoting effective interventions for neglected conditions in developing countries.** Disabil Rehabil; 29:973-976.
2. Olusanya BO, Newton VE. (2007). **Global burden of childhood hearing impairment and disease control priorities for developing countries.** Lancet; 369: 1314-1317.
3. Olusanya BO. (2007). **Addressing the global neglect of childhood hearing impairment in developing countries.** PLoS Med; 4(4):e74.
4. Olusanya BO. (2007). **"The right stuff": the global burden of disease.** PLoS Med; 4(2):e84.
5. Olusanya BO, Swanepoel de W, Chapchap MJ, Castillo S, Habib H, Mukari SZ, Martinez NV, Lin HC, McPherson B. (2007). **Progress towards early detection services for infants with hearing loss in developing countries.** BMC Health Serv Res; 7:14.
6. Olusanya BO. (2006). **Early hearing detection and intervention in developing countries: current status and prospects.** Volta Review; 106 [Monograph]: 381-418.
7. Olusanya BO, Roberts A. (2006). **Physician education and infant hearing loss in a developing country.** Pediatr Rehabil; 9:373-377.
8. Olusanya BO, Luxon LM, Wirz SL. (2006). **Ethical issues in screening for hearing impairment in newborns in developing countries.** J Med Ethics; 32: 588 – 591.
9. Olusanya BO, Eletu OB, Odusote B, Somefun AO, Olude O. (2006). **Early detection of infant hearing loss: current experiences of health professionals in a developing country.** Acta Paediatr; 95: 1300-1302.
10. Olusanya BO. (2006). **Health priorities for developing countries: importance of non-fatal but disabling conditions.** Trans R Soc Trop Med Hyg; 100: 1089-1090.
11. Olusanya BO, Okolo AA. (2006). **Adverse perinatal conditions in hearing-impaired children in a developing country.** Paediatr Perinat Epidemiol; 20:366-371.

12. Olusanya BO, Ruben RJ, Parving A. (2006). **Reducing the burden of communication disorders in the developing world: an opportunity for the millennium development project.** JAMA; 296: 441-444.
13. Olusanya BO, Okolo AA. (2006). **Revisiting the ten questions questionnaire for developing countries.** Int J Epdemiol; 35:1103.
14. Olusanya BO, Solanke OA, Okolo AA. (2006). **Stillbirths in sub-Saharan Africa.** Lancet; 368: 117.
15. Olusanya B, Mcpherson B, Swanepoel D, Shrivastav R, Chapchap M. (2006). **Globalization of infant hearing screening: the next challenge before JCIH?** J Am Acad Audiol; 17:293-295.
16. Olusanya BO, Okolo AA. (2006). **Early hearing detection at immunization clinics in developing countries.** Int J Pediatr Otorhinolaryngol; 70:1495-1498.
17. Olusanya B, Somefun A, Eletu O, Olude O, Odusote O. (2006). **Health professionals' readiness for infant hearing screening in Lagos, Nigeria.** East Afr Med J; 83:113-115.
18. Olusanya BO. (2006). **The burden of neonatal jaundice and sepsis in developing countries.** Trop Med Int Health; 11:381.
19. Olusanya BO. (2006). **Disabilities, physicians and the millennium development goals.** Disabil Rehabil; 28:244.
20. Somefun OA, Lesi FE, Danfulani MA, Olusanya BO. (2006). Communication disorders in Nigerian children. Int J Pediatr Otorhinolaryngol; 70:697-702.
21. Olusanya BO, Luxon LM, Wirz SL. (2006). **Maternal views on infant hearing loss in a developing country.** Int J Pediatr Otorhinolaryngol; 70:619-623.
22. Olusanya BO, Luxon LM, Wirz SL. (2006). **Screening for early childhood hearing loss in Nigeria.** J Med Screen; 12: 115-118.
23. Olusanya BO. (2005). **Can the world's infants with hearing loss wait?** Int J Pediatr Otorhinolaryngol; 69:735-738. Reprinted in *Audiology Today 2005; 17:10-11.*
24. Olusanya BO, Luxon LM, Wirz SL. (2006). **Childhood deafness poses problems in developing countries.** BMJ; 330:480-481.
25. Olusanya BO. (2005). **State of the world's children: life beyond survival** Arch Dis Child; 90:317-318.

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27. Olusanya BO, Luxon LM, Wirz SL. (2005). **Is negative parental attitude towards infant hearing screening justifiable?** Arch Dis Child; 90:546-549.
28. Olusanya BO. (2004). **Self-reported outcomes of aural rehabilitation in a developing country.** Int J Audiol; 43: 563- 571.
29. Olusanya BO, Luxon LM, Wirz SL. (2004). **Infant hearing screening: route to informed choice.** Arch Dis Child; 89:1039-1040.
30. Olusanya BO. (2004). **Polio-vaccination boycott in Nigeria** Lancet; 363:1912.
31. Olusanya BO, Luxon LM, Wirz SL. (2004). **Benefits and challenges of newborn hearing screening for developing countries.** Int J Pediatr Otorhinolaryngol; 68: 287 – 305.

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1. Olusanya BO, McPherson B. **Screening for hearing loss in developing countries.** In: Audiology for Developing Countries. McPherson & Brouillette, Editors. (in press).
2. Olusanya BO. (2006). **Early Hearing Detection in Developing Countries: A sound start for every child.** www.soundstart4all.com
3. Olusanya BO, Hodes D. (2005). **Between newborn and school hearing screening programmes.** Arch Dis Child [eLetter on Fonseca S, et al. School hearing screening programme in the UK: practice and performance. Arch Dis Child 2005; 90:154-156].
4. Olusanya BO. (2004). **When is early hearing intervention late?** Arch Dis Child. Foetal Neonatal Edition. [Letter on Kennedy C and McCann D. Universal neonatal hearing screening moving from evidence to practice. Arch Dis Child Foetal Neonatal Ed. 2004; 89:F378-F383].
5. Olusanya BO. (2005). **A Sound Start for Every Child**. Lagos. Touchstone Publishing, Lagos.

Presentations at Scientific Meetings

1. Olusanya BO. Priorities for early detection of childhood hearing impairment in developing countries. [Keynote Address]. 1st International Conference on Prevention and Rehabilitation of Hearing Impairment, Beijing, China, April 26-28, 2007.
2. Olusanya BO, Wirz SL, Luxon LM. Apgar Score as pre-screening tool for permanent hearing loss in resource-poor countries. 11th Spring Meeting of the Royal College of Paediatrics and Child Health, University of York, UK, March 26-29, 2007.
3. Olusanya BO, Luxon LM, Wirz SL. Infant hearing screening models for the early detection of permanent childhood hearing loss in Nigeria. IXth International Congress of the European Society of Pediatric Otorhinolaryngology, Paris, France. June 18-21, 2006.
4. Olusanya BO. Early hearing detection in infants in developing countries: options and challenges. [Keynote Address]. NHS 2006, Como, Italy, May 31 - June 3, 2006.
5. Olusanya BO, Luxon LM, Wirz SL: "Maternal Views on Infant Hearing Loss In Nigeria", 6th International Conference on Pediatric Otorhinolaryngology, European Society of Pediatric Otorhinolaryngology, Athens, Greece, May 16 – 19, 2004.
6. Olusanya BO, Luxon LM, Wirz SL: "Infant Hearing Screening in a Developing Country: Route to Informed Choice", 6th International Conference on Pediatric Otorhinolaryngology, European Society of Pediatric Otorhinolaryngology, Athens, Greece, May 16 – 19, 2004.

Other Conferences Attended

1. Early Hearing Detection and Intervention (EHDI) 2006 Conference, Salt Lake City, UT, USA. March 28-29, 2007.
2. Early Hearing Detection and Intervention (EHDI) 2006 Conference, Washington DC, USA. February 1 – 3, 2006.
3. XXVIIth International Congress of Audiology, Phoenix Az, USA. September 26 – 30, 2004.
4. Alexander Graham Bell Association for the Deaf and Hard of Hearing 2004 Convention, Anaheim, CA, USA. June 25 – 29, 2004.6